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# GELATINOUS MAMMARY CANCER

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ALTHOUGH rare, gelatinous cancer was among the first of the several varieties of mammary carcinoma to be described as a separate and distinct form. Its syrupy, jelly-like characteristics in the gross, and the peculiar ground substance under the microscope, make it readily recognizable and have permitted accurate classification. Lange[6] has credited Otto,[9] in 1816, with the first description of gelatinous carcinoma. Robinson,[11] in reporting a case to the pathologic Society of London, in 1852, stated that only four similar tumors affecting the breast had been recorded, one by Jonathan Müller,[8] one by Rokitansky,[12] one in the London Hospital Museum, and one in St. Thomas's Hospital.

The history of Robinson's case is of interest in that the patient remained free of the disease for five and one-half years after excision. He related that the patient, age 56, "was under the care of the late Mr. Tyrell seven years ago (1827) for a tumor (not ulcerated) in the right breast, and another in the right armpit, both of which were removed by the knife and the wound healed perfectly in a month. . . . About 18 months ago, a hardness was noticed on the termination of the wound towards the nipple. . . . About one-third from the outer termination of the cicatrix is a small lobulated tubercle, which has existed four months, and which evidently increased. It is of purple colour, covered with crimson vessels, and surrounded by a similar stony induration, firmly attaching it to the chest.

"November 27, (1834): The ulcers had scarcely varied at all; the redness of the integuments of the right arm and the swelling somewhat subsided, but the hardness and tenseness continued. She had a troublesome cough, wheezing, dyspnoea and sense of weight in the chest, with great emaciation; and although relieved by blistering, expectorants, hyoscyamus, and mild tonics, she died in the evening from effusion into the chest. . . . At autopsy . . . the tumors in the course of the cicatrix had extended as far as the ribs, and implicated them. They were specimens of colloid cancer, and appeared much the same as during life, although not so vascular. One had a fibrous appearance, the other, a vascular; and into one of these blood had been effused. Upon incising it posteriorly, the gelatiniform appearance was beautifully

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shown. The lungs were congested, and the air-cells somewhat dilated, but they were not at all affected by malignant disease. There was about a pint of serous fluid in each side of the chest. The heart was healthy. The other viscera were not examined."

Gaabe,[3] in 1908, found that this type of cancer was twice as curable as the ordinary form. Halsted,[4] in 1915, described a peculiar sensation on palpation which he thought was diagnostic of the gelatinous cancer. He stated that the tactile impression "might be defined as a delicate swish or crush of a jelly-like structure under tension." Lee, Hauser and Pack[7] reviewed the

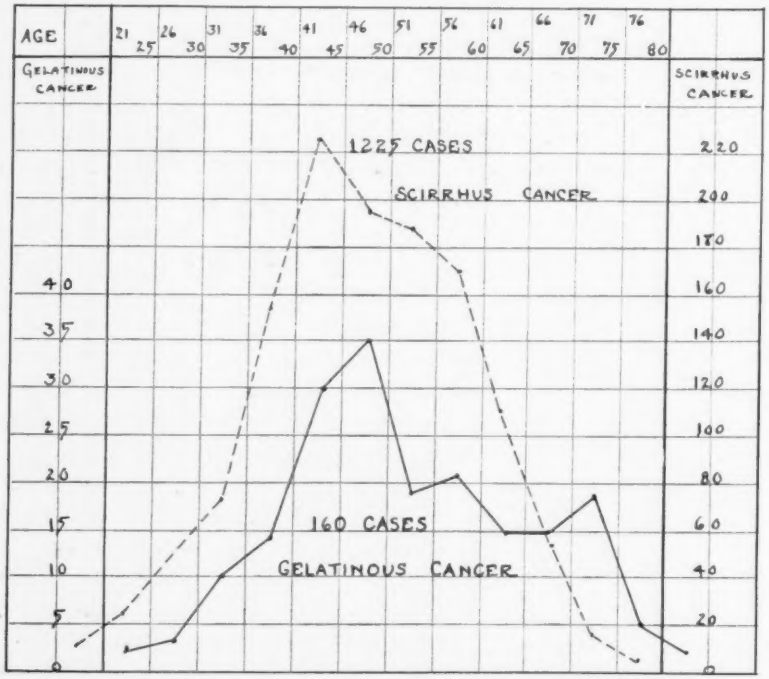



CHART 1.—Shows the comparison of the age incidence of 1,225 cases of scirrhous cancer with 160 cases of gelatinous cancer (including the author's series and those collected by Gaabe).

literature on the subject, in 1934, and found the incidence given by various authors as 1 to 2 per cent of all cancers of the breast. They reported 30 cases, recorded in the Memorial Hospital, with 57 per cent of five-year cures.

The present study is based upon 83 cases of gelatinous carcinoma recorded among a total of 2,300 cancers of the breast in the Surgical Pathological Laboratory. This study emphasizes the slow growth of these tumors, their low degree of malignancy and their tendency to local recurrence five to 30 years after treatment by radical operation.

*Clinical Features.*—Gelatinous carcinoma occurs on the average in more elderly patients than ordinary mammary cancer. One-third of the cases in the present series occurred between the ages of 41 and 50 and nearly an equal number occurred in patients between the ages of 60 and 79. The peak of the age incidence was between 46 and 50 years compared to a peak between

41 and 45 years for ordinary cancer of the breast (Chart 1). The average age in the series of Lee, Hauser and Pack was 52 years.

A discovery of a lump in the breast was the first sign noted by the patient in 80 per cent of our cases. The painless character of the lump and its slow growth enhance the natural period of delay in consulting a physician for treatment. In 33 cases, the lump was known to be present for a year or more before the first examination and in nine of these cases, the duration of the lump was five years or longer. In 36 cases, the duration of the lump was less than a year, the average duration in this group being 4.1 months compared to 4.3 years for the group in which the duration of symptoms was a year or more. In three instances, the lump was discovered during routine examination, the patient being unaware of its presence. One patient had noticed hardness of the affected breast for 20 years, the hardness apparently disappeared for several years but recurred eight years before admission and rapidly increased in the past year. During the past three years she had had a sanguineous discharge from the nipple. When examined, in 1915, there was diffuse induration of the entire breast with a soft fluctuating mass 3 cm. in diameter beneath the nipple. This patient died six years after radical operation with invasion of the mediastinum.

Gaabe has estimated (on the basis of 88 cases collected from the literature) that the duration of symptoms in gelatinous carcinoma is two and one-half times that of the more common scirrhus form. Lange, basing his conclusions on a series of breast cancers reported by various German authors, states that ulceration of the skin in scirrhus cancer occurs from one to two years after the onset of symptoms. In the 75 cases of gelatinous carcinoma collected by him, the average period elapsing before fixation of the skin was 33.9 months and ulceration occurred after an average period of 57.7 months.

The statement by patients, giving the duration of their tumor in terms of years, is corroborated by the similar period which has elapsed between local excision and the recurrence of a tumor of appreciable size in the scar. In the case reported by Robinson, cited above, the first signs of local recurrence were five and one-half years after excision. In a similar case, recorded in this laboratory, local excision was performed in October, 1919, for a tumor the size of a walnut, thought to be a retention cyst. The recurrent tumor, which was half again as large, was noted in the breast in February, 1922. A simple amputation of the breast was performed, and, in November, 1930, an enlarged axillary lymph node was excised which showed mucoid cancer. The patient was reported well in April, 1937. Halsted performed excisions for recurrent tumors, in 1903 and 1906, for a case which had had two previous excisions at four-year intervals. This patient died in an automobile accident in 1931, at the age of 73.

Lee, Hauser and Pack have emphasized the large size of some of these growths. Lange found that the tumors varied in size from a walnut to that of an orange. Only two of his cases were the size of a child's head. Gaabe found that 10 per cent of 88 cases were of unusual size. The present study

emphasizes the relatively small size of the tumor in comparison with the long duration of symptoms. The average size of the tumor in 58 cases was 5.3 cm., and approximately 50 per cent had a diameter of 4 cm. or less. There were only five tumors, 10 cm. or over in diameter, the largest, 22 cm., was a recurrence after local excision. Three cases had the diameter of 8 cm., and one, 9 cm. (Chart 2).

Beside its slow rate of growth, and the tendency for the tumor to occur near or after the menopause, there is very little additional information that

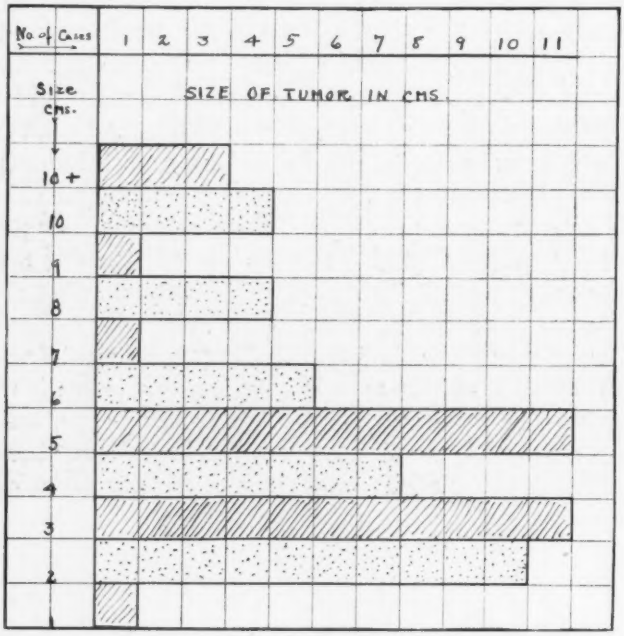



CHART 2.—Shows the maximum diameter of the tumor in cases of gelatinous carcinoma.

can be gleaned from the histories given by these patients. Pain is usually not an early symptom. Approximately 10 per cent of the patients noted a discharge from the nipple, usually of a sanguineous character, and an approximately equal number had noted change in the size or color of the nipple, with or without itching or irritation. The history of trauma or preceding lactation mastitis occurred in an insignificant number. Discoloration or ulceration of the skin prior to examination was noted in 14 cases. Where present, this was usually given as the reason for seeking advice. In general the skin changes were present only in those tumors which were six or more centimeters in diameter. There was one exception.

There are four chief findings on clinical examination which suggest a diagnosis of gelatinous carcinoma. The first is the relatively small size of the tumor in comparison with the long duration of symptoms. The second is the protrusion and enlargement of the nipple on the affected side. This finding of protrusion, rather than retraction, was noted by the examiner in eight cases in the present series and the finding is probably of more frequent occurrence, since its diagnostic importance in gelatinous carcinoma

has not been heretofore emphasized (Fig. 1). The third is the cystic character of the growth on palpation. The diagnosis of benign cyst was made prior to operation in 11 cases in the present series.

The differentiation from a benign cyst should not be difficult in these cancers because the tumors are often larger (4 to 6 cm.), and on aspiration do not yield the cloudy fluid obtained from a cyst of similar size. They are accompanied by atrophy of the overlying fat. The fourth diagnostic finding

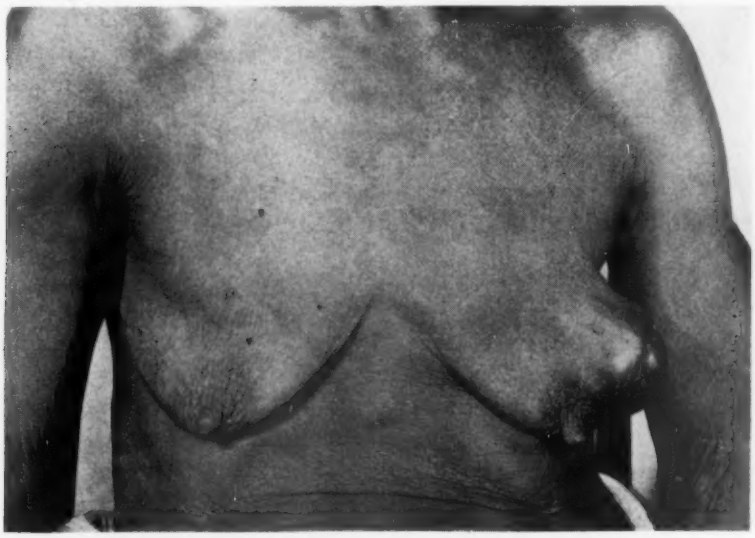


FIG. 1.—Path. No. 60479: Gelatinous carcinoma occurring in a colored woman, age 62. Note the bulging and enlargement of the nipple on the affected side. Enlargement and protrusion of the nipple may be a diagnostic sign in these growths.

in the present series, is the feeling of a "swish" on firm pressure, originally described by Halsted. This impression of a ruptured cyst, or the finding of an area of fluctuation in an otherwise solid tumor are important in diagnosis.

Gelatinous carcinoma occurs most frequently in the outer upper quadrant and in the central portions of the breast. The upper inner quadrant, the lower outer quadrant, and the lower inner quadrant are affected in the order of frequency given. The location of the tumors studied in the present series and those reported by Lee, Hauser and Pack and by Gaabe, are shown in Table I.

TABLE I

LOCATION OF THE TUMOR IN 91 INSTANCES OF GELATINOUS CARCINOMATA

| Author | Upper Outer Quadrant | Upper Inner Quadrant | Center | Lower Outer Quadrant | Lower Inner Quadrant | Entire |
|---|---|---|---|---|---|---|
| Author's series | 11 | 8 | 16 | 5 | 1 | 4 |
| Lee, Hauser and Pack | 4 | 4 | 10 | 0 | 3 | 1 |
| Gaabe | 15 | 3 | 0 | 6 | 0 | 0 |
| Totals | 30 | 15 | 26 | 11 | 4 | 5 |

In seven of the cases in the present series, there were multiple tumors in the same breast; these were either in the outer or upper portions of the breast. In three cases both breasts were ultimately involved. In one, the involvement of the second breast occurred one year after the radical operation for the first tumor and was a mucoid cancer. In the second case, the involvement three years later showed a scirrhus cancer with mucoid change. In the third case, a similar tumor appeared beneath the nipple in the opposite breast and enlarged nodes were palpable in the axilla five years after radical operation. In the 75 cases studied by Lange, three developed cancer in the other breast. In one, the tumor in the second breast was a colloid cancer. The second was diagnosed as scirrhus and the third as Paget's disease.

*Differential Diagnosis.*—While the diagnosis of gelatinous cancer is usually made on the gross specimen or the microscopic section, there are 11 cases in the present series in which the nature of the tumor was correctly inferred from the clinical findings. The relatively small size of the tumor in cases where the symptoms had been present several years and its encapsulated rounded character with a boggy or fluctuant feeling, together with atrophy of the overlying fat, dimpling, or palpable axillary lymph nodes, suggested the presence of mucoid carcinoma. In four of the five cases reported by Halsted, and included in this series, the impression of rupture of delicate membranes or "swish" led to a diagnosis of mucoid carcinoma.

As mentioned above, the tumor was incorrectly diagnosed as a benign cyst in 11 instances. In four cases, the impression was that of an intracanalicular myxoma. In an additional four cases, the diagnosis was benign intracystic papilloma or papillary carcinoma. In the remaining cases, where the diagnosis of gelatinous carcinoma was not made clinically, the clinical impression was that of ordinary mammary cancer. In the cases where the diagnosis of benign cyst was made, sufficient importance was not given to the nearness of the tumor to the skin, the atrophy of the overlying fat, its size, or its failure to yield a characteristic cloudy fluid on aspiration. The distinction between a giant intracanalicular myxoma, or degenerating fibro-adenoma is more difficult. Usually the fibro-adenomata of corresponding size are opaque on transillumination, whereas in gelatinous carcinoma if the lesion is soft and sufficiently gelatinous the tumor will transilluminate. If a large size (No. 18 gauge) needle is used for aspiration, the characteristic gelatinous material should be demonstrable (Fig. 2).

Papillary adenocarcinoma or benign intracystic papilloma may be simulated by these growths. A central location, a sanguineous discharge from the nipple, and a boggy feeling with or without areas of fluctuation, occur in both gelatinous and papillary cancer. That the differential diagnosis may be impossible clinically, is indicated by the pathologic finding of mucoid change in eight cases of papillary cancer recorded in this laboratory.

*Gross Pathology.*—Careful study of the gross and microscopic pathology of mucoid carcinoma indicates that there are two groups of mammary cancers which contain this type of gelatinous material. In one group, the tumors

contain a shiny, translucent gel throughout their substance, in the second group, the gelatinous change is found in one or more portions and solid, dense tissue in the remainder of the growth. In the present series, 59 tumors showed a characteristic gelatinous substance throughout, and 24 showed secondary mucoid change in cancers of the scirrhus or duct type.

The gross specimens of typical gelatinous cancer may be cystic or solid, encapsulated or nonencapsulated. The group of mammary cancer in which mucoid change is found in isolated portions of the gross specimen only, or under the microscope, have shown grossly a resemblance to scirrhus or duct cancer.

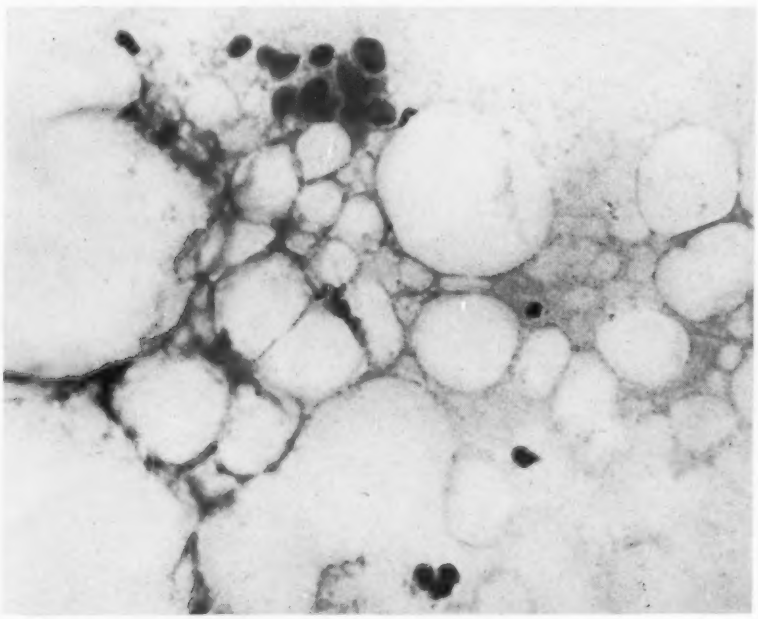


FIG. 2.—Path. No. 69609: Photomicrograph showing cancer cells in the mucoid material aspirated from a case of gelatinous carcinoma.

In those of carcinoma with complete gelatinous change, 16 of the cases were described as encapsulated and two as circumscribed. The encapsulated tumors at exploration resemble a cyst or degenerated intracanalicular myxoma. In one instance, the cyst-like tumor presented a blue dome because of its vascularity. In another the tumor was polycystic. When the capsule of these cystic tumors is incised the soft, translucent jelly-like material, often flecked with hemorrhage, bulges or flows from the incision. The exuded material has frequently been compared to tapioca (Figs. 3 and 4).

Encapsulated gelatinous carcinoma may closely resemble degenerating intracanalicular myxoma grossly. However, in the true myxomata the mucoid tissue is firm, and does not exude from the cut surface.

In the nonencapsulated gelatinous carcinomata, the mucoid material may invade the surrounding tissues, including fat and muscle, or the gelatinous

material may be diffuse throughout the more solid growth giving the cut surface a slimy, grayish cast or a honey-combed appearance (Figs. 5 and 6). Three of the nonencapsulated tumors were fixed to the overlying skin and two had invaded the pectoralis muscle. In such solid nonencapsulated tumors, pockets of mucoid material such as are found in encapsulated tumors may occur (Fig. 7).

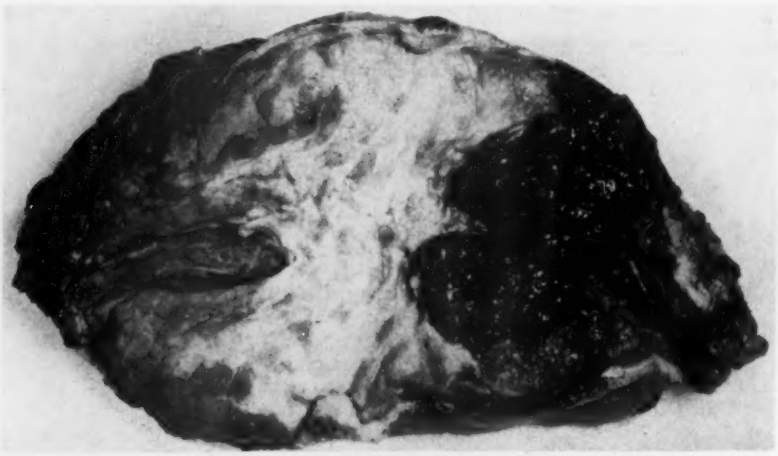


FIG. 3.—Path. No. 24144: Photograph of a gross specimen showing the bulging dark jelly-like material in a case of encapsulated gelatinous carcinoma.

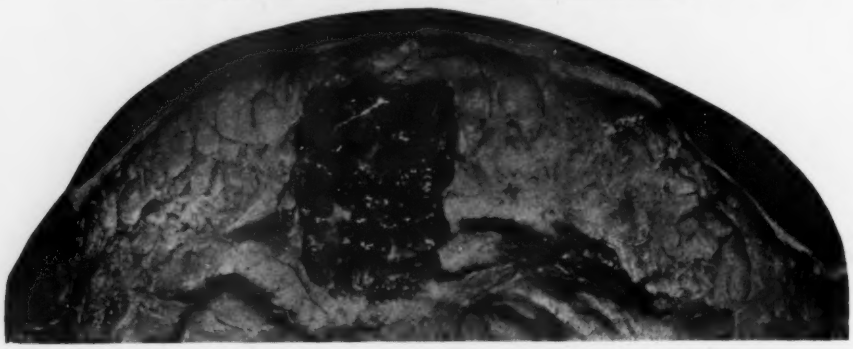


FIG. 4.—Path. No. 4874: Photograph of a gross specimen showing the encapsulated character of gelatinous growth, which feels like a cyst when palpated.

In six cases in the present series, gelatinous material was interspersed in tumors having the structure of papillary carcinoma grossly (Fig. 8). This type of gelatinous carcinoma has been well described and illustrated by Lee, Hauser and Pack. In two other cases, the papillary carcinoma showed a typical structure and pockets of mucoid material were found in isolated portions of the tumor (Fig. 9). More frequently, when only partial mucoid change is found, the tumor is a slowly growing scirrhus carcinoma of large size (Figs. 10A and B).

*Microscopic Pathology.*—Gelatinous change has been described in nearly all varieties of mammary cancer. In general, the more slowly growing forms

of carcinoma of the breast are more prone to mucoid change, but this type of gelatinous material may be found in isolated portions of more malignant forms. In such cases the gel is apparently deposited in foci where interruption of blood supply, or other factors have interfered with growth. Gelatinous carcinoma, therefore, can be divided microscopically into those cases

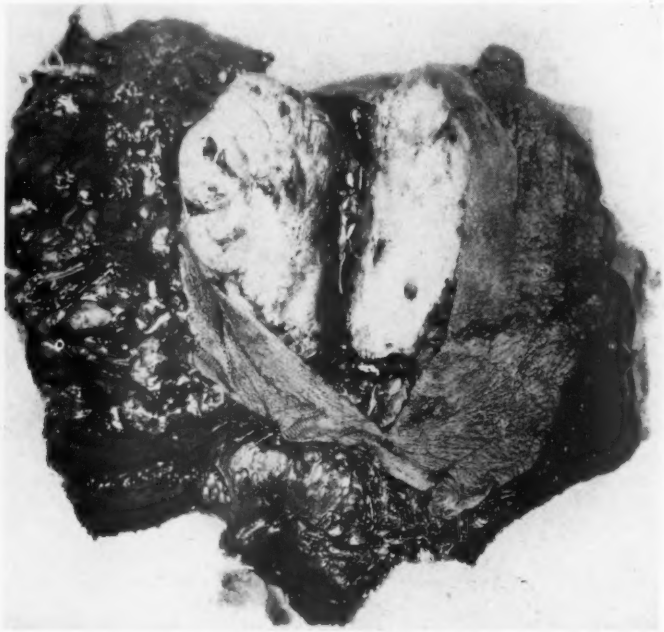


FIG. 5.—Path. No. 60479: Gelatinous carcinoma of solid character showing a characteristic glary surface.

FIG. 6.—Path. No. 17869: A solid gelatinous growth removed at operation for recurrence, 19 years after mastectomy for the primary tumor.

in which the gelatinous material predominates throughout the entire tumor and cases in which the gelatinous material is deposited in isolated foci or sparsely through solid tumor growth. The majority of gelatinous carcinomata contain epithelium with definitely malignant features, resembling that found in

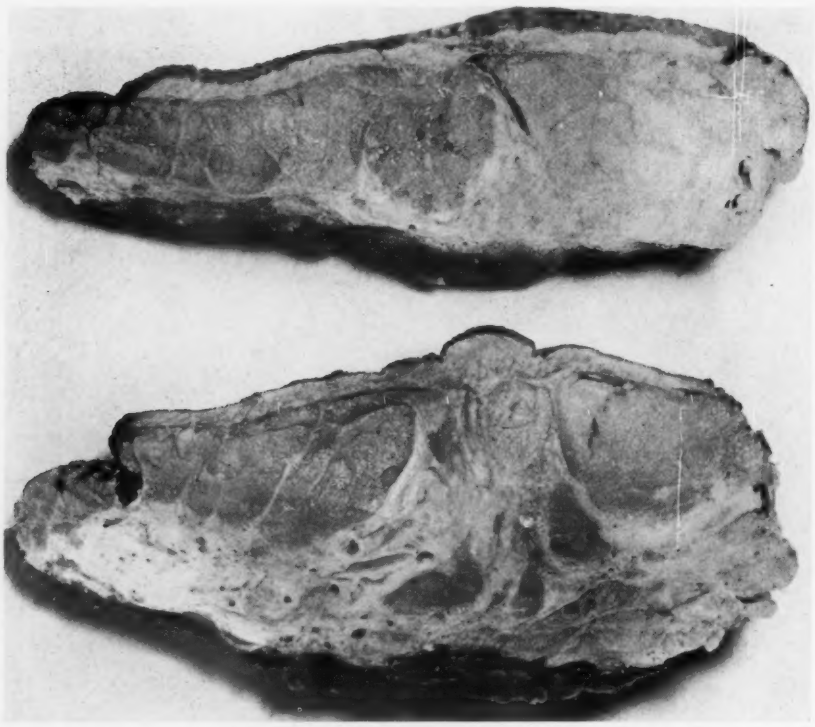


FIG. 7.—Path. No. 29697: A specimen of gelatinous carcinoma combining cystic and solid features.

FIG. 8.—Path. No. 9733: A specimen showing mucoid change in a papillary carcinoma (so-called, bulky cystadenocarcinoma).

ordinary scirrhus or papillary cancer, whether or not the mucoid substance is diffuse and abundant or scarce (Fig. 11).

Where the cancer cells resemble those found in ordinary scirrhus carcinoma, the degree of malignancy and the prognosis vary with the amount of mucoid substance found. This is in contrast to the view of Cheatle and Cutler,[1] who stated that the clinical course does not depend upon either the presence or the extent of the gelatinous degeneration. In the present series, there were 58 cases with malignant epithelium of the scirrhus type. In 34 of these cases, the gelatinous change was diffuse throughout the tumor and the percentage of five-year cures was 54. In 24 cases, mucoid change appeared in isolated portions of the growth or was exceedingly scarce. In

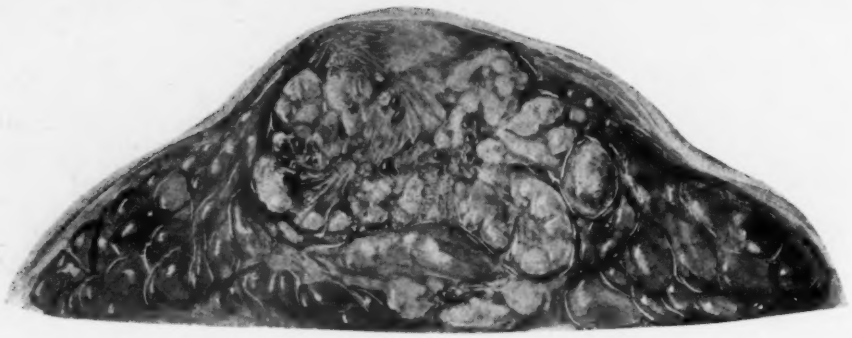
FIG. 9.—Path. No. 50573: Papillary carcinoma showing mucoid change. The papillary structure is seen at the upper margin of the growth and the gray gelatinous material at the lower. There is an intervening dark zone of hemorrhage.

this latter group, there were 30 per cent of five-year cures, a percentage of survivals which was identical with that found in 1,225 cases of ordinary scirrhus carcinoma recorded in this laboratory.

Among the carcinomata of the breast in which gelatinous material predominates, there is a third group of cases containing epithelium of the basal cell type. Judging from the relatively benign character of the epithelium and from the large amount of mucoid substance, the prognosis in these growths should be unusually good. This assumption is borne out by the cases of this character in this series. Nineteen of 25 cases were traced and, with the exception of three dying from other causes, all survived the five-year period (100 per cent of five-year cures).

Among those gelatinous carcinomata with cells approaching the basal cell type, there is a group which is microscopically indistinguishable from basal cell cancer of the adenocystic type, such as is found in the parotid gland,

A



B

FIGS. 10A and B.—Path. Nos. 10639 and 7567: Gross specimens of slowly growing scirrhus carcinoma with areas of gelatinous degeneration.

TABLE II

AN ANALYSIS OF THE FIVE-YEAR MORTALITY RATE BASED UPON THE TYPE OF CELL

| Type of Cancer | No. of Cases | Not Traced | Dead Within 5 Years | Dead Other Causes | Dead After 5 Years | Well Over 5 Years | Per Cent 5-Year Cures |
|---|---|---|---|---|---|---|---|
| Basal cell cancer, with complete mucoid change... | 25 | 6 | 0 | 3* | 3 | 13 | 100 |
| Papillary or scirrhus cancer, with complete mucoid change..... | 34 | 8 | 11 | 2 | 4 | 9 | 54 |
| Scirrhus cancer, with partial mucoid change.... | 24 | 4 | 13 | 1 | 3 | 3 | 30 |

* One of these patients died postoperatively.

and a portion of the growth may resemble benign mixed tumors of the parotid (Fig. 12). That typical mucoid carcinoma of the mammary gland may originate as a form of adenocystic basal cell cancer, is suggested by three cancers of the breast, recorded below, in which the mucoid change at operation was not yet pronounced and the predominating structure was adenocystic basal cell cancer.

**Path. No. 56134.**—This case was referred to the laboratory by Dr. Louisa A. Keasbey of Lancaster, Pa. The patient is a white female, age 46. There is a small lump in the right breast which was noted for one year. The patient has had three normal pregnancies, the last 23 years ago. She still menstruates regularly. There are no other

FIG. 11. FIG. 12.

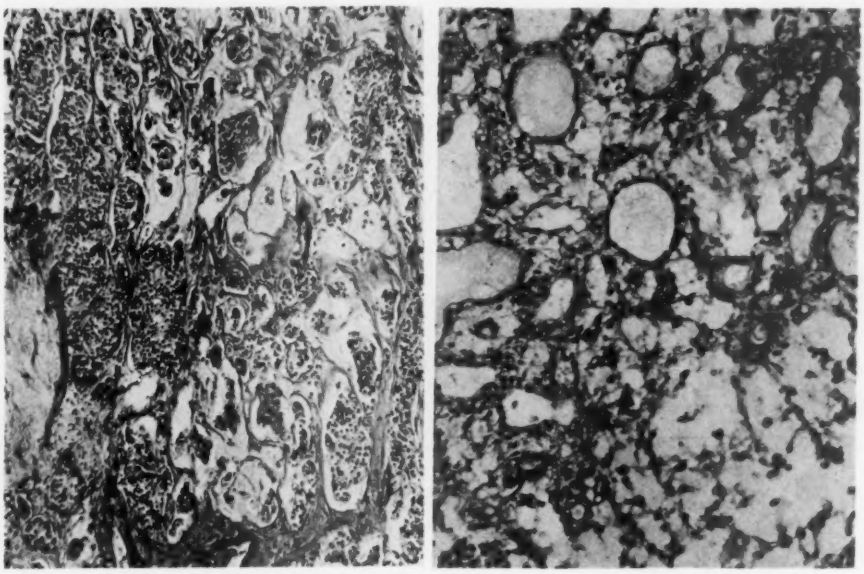


FIG. 11.—Path. No. 41029: Photomicrograph of scirrhous carcinoma with mucoid change. The majority of gelatinous carcinomata show cancer cells of this type but with more abundant gelatinous material.

FIG. 12.—Path. No. 27946: Photomicrograph of a gelatinous carcinoma resembling a mixed tumor of the parotid gland. This patient remained well 14 years following radical operation.

abnormalities in either breast. The tumor, which is well circumscribed and situated deeply in the breast tissue, is 3.5 cm. in diameter. A radical mastectomy was performed in February, 1935. The tumor was definitely circumscribed and contained one small cyst, 0.5 cm. in diameter.

Microscopically, the tumor has the appearance of adenocystic basal cell cancer. There are numerous acini of variable size containing a mucoid secretion (Fig. 13). The patient was reported well in September, 1937.

**Path. No. 35154.**—The patient was a woman, age 44. The tumor, 2 cm. in diameter, was not adherent to the skin or deep structures, and was situated in the left breast. There were no palpable nodes in the axilla. The upper half of the breast only was removed down to the pectoral muscle in April, 1924. Dr. Bloodgood reported on the section as probable low grade carcinoma and classified it as borderline but recommended that it be treated as cancer (Fig. 14). The patient was given instead postoperative irradiation. In December, 1924, she had had a goiter operation, additional roentgenotherapy, and nodules were palpable in the other breast. There is no note of additional therapy and the patient was reported apparently well, May 12, 1930.

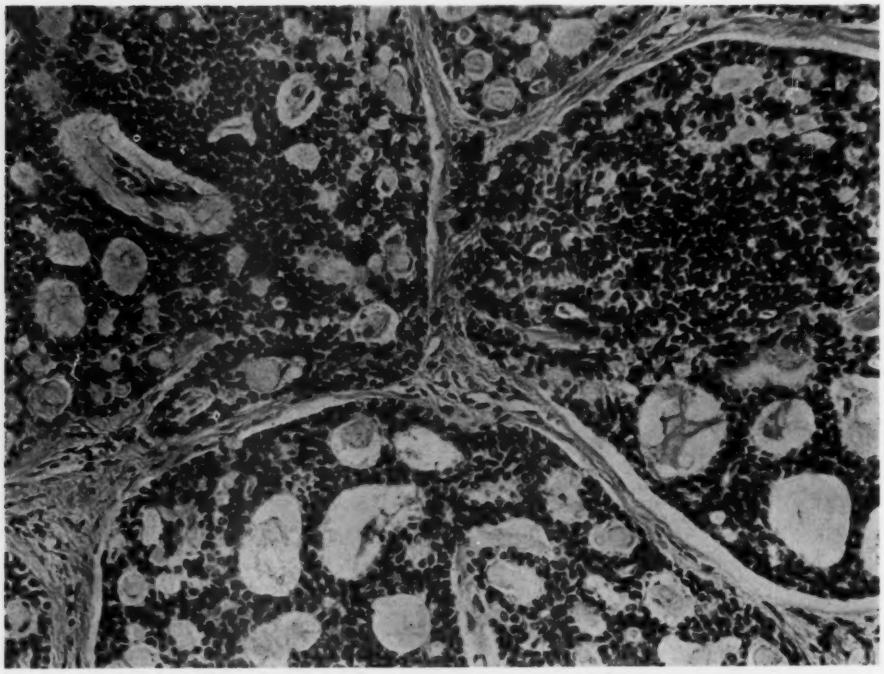


FIG. 13.—Path. No. 56134: Photomicrograph of an adenocystic basal cell carcinoma of the breast with areas of mucoid secretion (case of Dr. L. A. Keasbey, Lancaster, Pa.).

FIG. 14

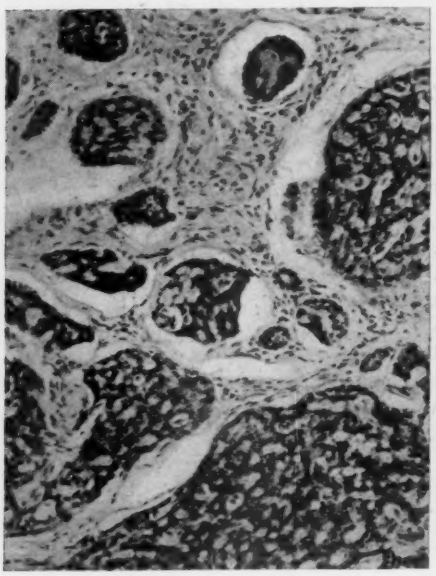


FIG. 15

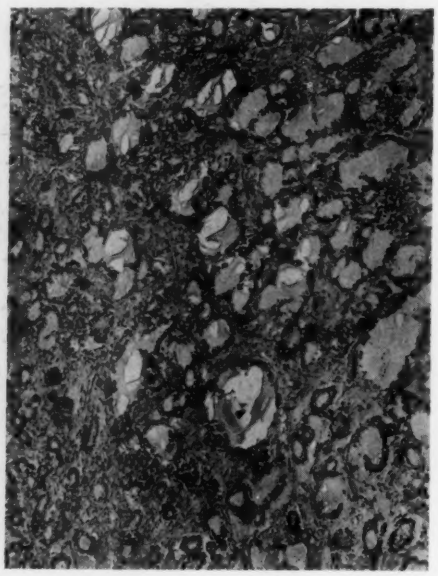


FIG. 14.—Path. No. 35154: Photomicrograph of an adenocystic basal cell carcinoma of the breast. The tumor was treated by excision and postoperative irradiation. The patient has remained well six years.

FIG. 15.—Path. No. 39095: Photomicrograph of a basal cell cancer of the breast containing tissue suggestive of benign adenoma of the parotid gland. Note the dilated acini with mucoid secretion.

**Path. No. 39095.**—The patient is a white woman, age 46, who had a tumor excised from her breast elsewhere. The specimen contains an encapsulated tumor mass 5 cm. in diameter. The capsule is intact at every point and the mass is embedded in fat. The tumor has a soft and spongy feeling and on section it is a slaty-black color. The gross diagnosis suggests hemangioma.

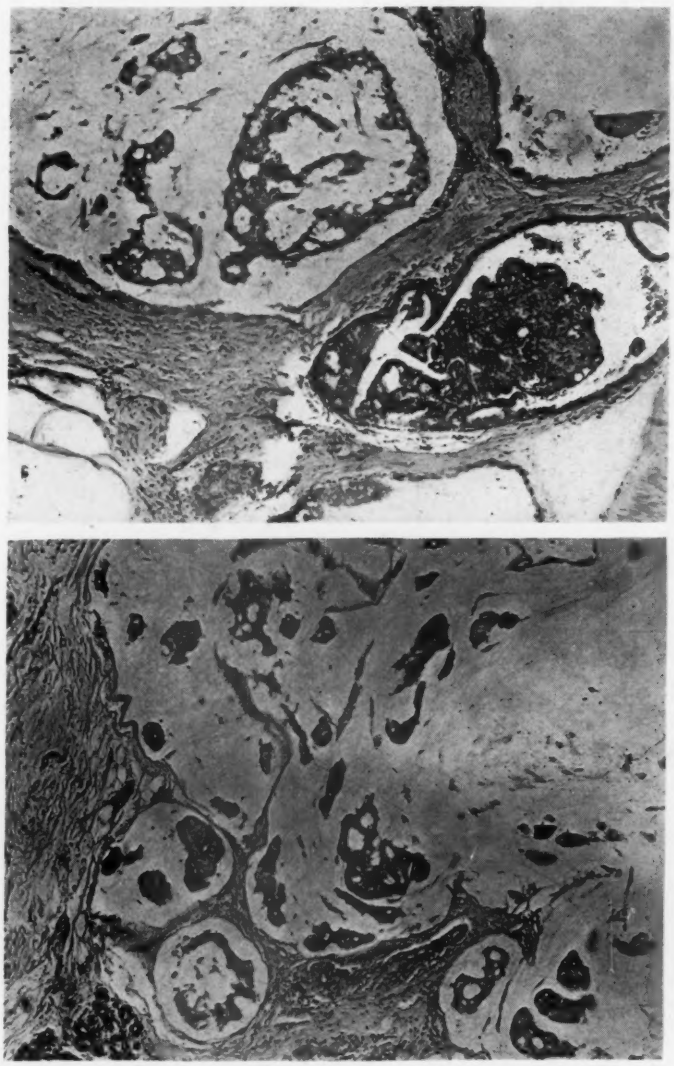


FIG. 16.—Path. No. 35024: Photomicrographs of a small papillary tumor of the breast undergoing mucoid change. The patient, age 30, had two small tumors about 1 cm. in diameter, one near the right and one near the left nipple. The nodules were treated by simple excision. The patient has remained well 14 years without further treatment. This case illustrates the origin of mucoid carcinoma from papillary tumor and the low degree of malignancy of such tumors in their early stage of development.

The section of the tumor shows strands of basal cells which differentiate toward cuboidal cells, forming definite acini enclosing large amounts of colloid-like material (Fig. 15). The coagulated material extends into the stroma and many of the acini are

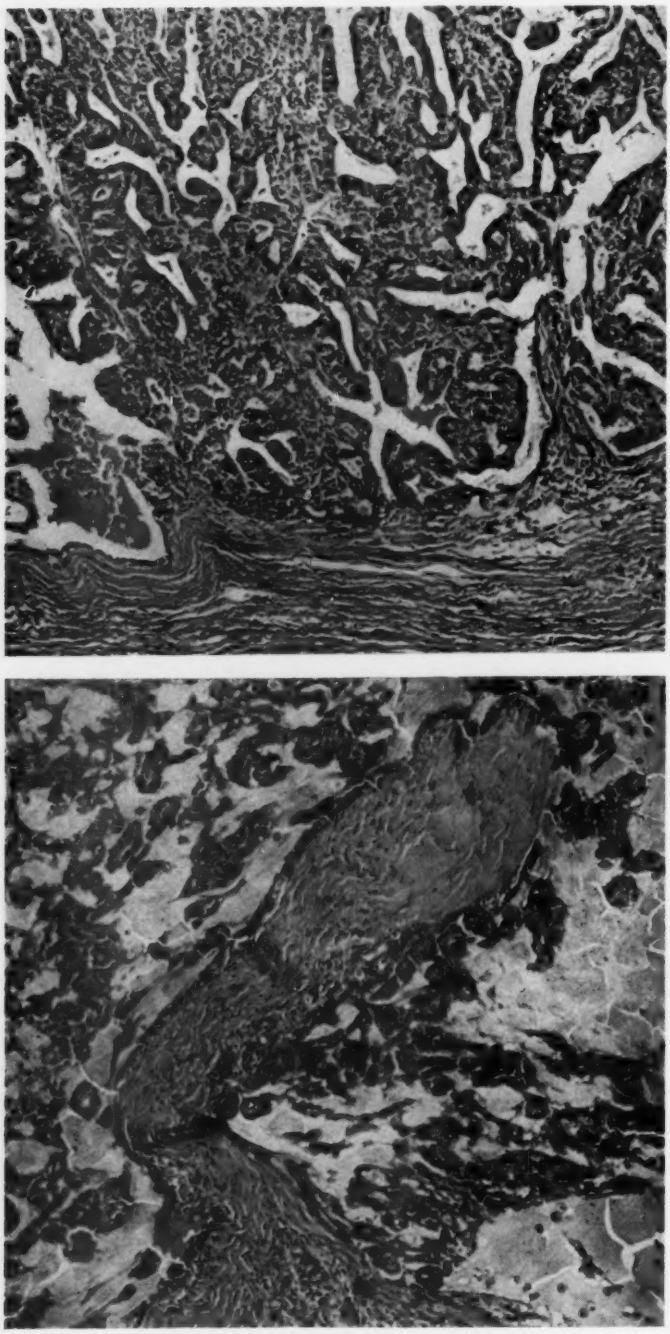


FIG. 17.—Path. No. 50507: Photomicrographs of a papillary carcinoma with mucoid change. This patient was operated upon four times, over a period of 30 years. She has remained well six years since the last operation. The upper picture shows the papillary structure and the lower the gelatinous portion.

ruptured. The tumor has a definite fibrous capsule, the surrounding lobules of breast are atrophic and embedded in loose periductal fibrous tissue which stains poorly. At one point in the surrounding breast tissue bordering on the tumor there is a duct adenoma. None of the tumor cells have a definitely malignant appearance, although there is a moderate variation in the size and density of the nuclei.

Mammary carcinoma with large amounts of gelatinous material infiltrating the entire tumor may contain islands of tumor cells suggesting an origin from slowly growing scirrhus or papillary cancer. Such tumors contain small alveolar structures with malignant cuboidal cells surrounded by a characteristic gel, encased in the remnants of periductal or perilobular connective tissue. Such slowly growing adenocarcinomata may contain portions indicating an origin from benign or malignant papillomata (Fig. 16). More often the relation to papillary carcinoma is indicated by the gross specimen (Figs. 10 and 11). Three cases of papillary carcinoma with partial mucoid change have been excluded from the present series because of relative insignificant amounts of mucoid material. In the following case mucoid change predominated in the recurrent tumors.

**Case Report.**—Path. No. 50507: The patient at the age of 18, in 1902, had a benign papilloma removed from the right breast with a good margin of breast tissue. Her first pregnancy was at the age of 27, during the next six years she had four more children. There were recurrent swellings in the right axillary region with each pregnancy which spontaneously regressed during lactation.

Eight years after the birth of her last child and 23 years after the first operation, at the age of 41, a small nodule appeared in the scar. This nodule gradually enlarged, the overlying skin showed a purple discoloration, broke down, and discharged a dark, bloody fluid. At a second operation, in 1925, this mass, which proved to be a papillary carcinoma, was removed and a dissection of the nodes of the right axilla performed. Four years after the second operation, nodules appeared in the scar in the region of the axillary prolongation of the incision. These enlarged slowly and became tender during the next two years. At the age of 47, in November, 1931, 29 years after the first excision, a third operation was performed and the recurrent masses removed. At this operation gelatinous material flowed from the wound. During the next ten months additional masses appeared in the right axilla and a fourth excision was performed in October, 1932. The patient was reported well five and one-half years later, in April, 1938.

The tissue from the first operation was reported as a benign intracystic papilloma. That from the three subsequent operations showed papillary carcinoma with mucoid degeneration (Figs. 9 and 17).

In most of the cases recorded in this series, a form of scirrhus carcinoma with a tendency to differentiate alveolar-like structures was apparently a starting point of the gelatinous tumor. The more definite the alveolar-like structures the greater tendency to mucoid change. Where the histology approached typical scirrhus carcinoma, the amounts of gelatinous material were small or were found only in isolated portions of the tumor.

*Origin of the Gel.*—The present study indicates that a variety of mammary carcinomata may undergo mucoid changes, resulting in typical gelatinous carcinoma. Slowly growing scirrhus cancer, papillary adenocarcinoma,

and adenocystic basal cell cancer are the most frequent sources for growths in which the characteristic gelatinous material pervades the entire tumor structure. Kaufman,[5] Lange[6] and Ewing[2] are among those authors who have sought to trace the origin of the gelatinous material to stromal degeneration. The abundance of the gel and the crowding out of epithelial elements in some of these growths favors such an interpretation (Fig. 18). The gelatinous material, moreover, does not take the specific stains for mucin. Ribbert,[10] Gaabe,[3] and most recent contributions hold that the gel results from the secretory ac-

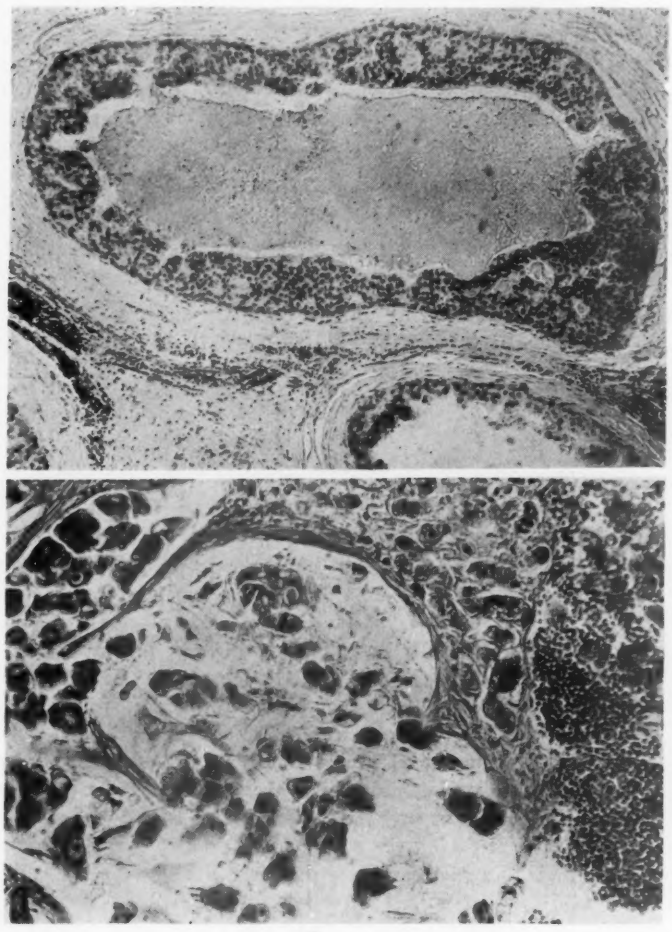
FIG. 18.—Path. No. 18441: Photomicrograph of gelatinous carcinoma where the mucoid change is crowding out the epithelial elements which resemble basal cells.

tivity of the malignant epithelium. There is a preponderance of evidence in support of such a view. The occurrence of the gel in metastatic deposits in the lung and lymph nodes, where fat and fibrous material are sparse or absent, indicates an epithelial origin (Fig. 19). Moreover, in papillary tumors and adenocystic basal cell cancers of the breast undergoing mucoid change the gel can be seen forming within the acinar structures of the tumor away from stromal elements (Figs. 12 and 13).

*Treatment.*—A study of the results of treatment in gelatinous carcinoma emphasizes the tendency to late recurrence and a long survival period, in spite of incomplete or unsuccessful surgery. A period of 5 to 10 years of freedom

from symptoms following treatment does not necessarily indicate permanent cure. In the present series, there were 14 cases who suffered late recurrences. Three patients died in the sixth year following radical surgery, two in the seventh, one in the eighth year, and one in the tenth, and another had a similar cancer in the opposite breast after five years. Two patients died of

A



B

FIG. 19 A and B.—Path. No. 8030: Photomicrograph of comedo carcinoma in which gelatinous change occurred in the metastatic nodules of the lymph node but not in the primary growth. Above is illustrated the structure of the primary growth; below cancer cells with mucoid secretion in the metastasis to the lymph node.

metastases, 16 and 18 years, respectively, after the complete operation, and a third had metastases 14 years later. Two patients had repeated excisions for the disease over a period of 11 years, one remaining well eight and the other 25 years after the last operation. Another patient had three operations

in seven years and is well six years after the last excision. In all, 14 patients survived the last treatment by 11 or more years, and among these 11 have remained well.

Of 83 gelatinous cancers, 65 have been adequately traced and of these 40 or 61 per cent survived the five-year period, the remaining 25 dying of the disease. If the cases with diffuse gelatinous change, discovered only after microscopic study, are included, there are 76 per cent of five-year cures, and if among these only, those showing basal cells are considered, the five-year survivals are 100 per cent.

In 19 cases, the initial treatment was excision only, in one no treatment was given, and in five cases simple mastectomy was performed. In 12 of the 19 cases, where excision was employed, the radical operation was performed in the next few days. The remaining cases were treated by further partial operations (simple mastectomy and excisions) or by deep roentgenotherapy. In five cases, simple mastectomy was the initial treatment, followed by further surgery in only one instance.

No cures were established by simple excision alone with the exception of the case illustrated in Fig. 16. One case treated in addition by deep roentgenotherapy is living six years. One case was treated by excision in 1919, by simple mastectomy in 1922, and by excision of diseased axillary nodes in 1930, and is well in 1938. Another patient was treated by excision in 1895 and 1899; received further dissections by Halsted in 1903 and 1906, and died from accidental death in 1931. One patient treated by simple mastectomy was well 16 years later and another is well after six years. The history of two cases initially treated by incomplete operation who survived for more than 15 years, but eventually succumbed to the disease, is given below.

**Path. No. 26200.**—The patient, a colored female, age 40, was treated for enlargement of the thyroid with iodine, 12 years ago. Four and one-half years ago, she noticed a small lump in the upper outer quadrant of the left breast. This increased gradually to the size of a walnut. She struck it with a piece of wood after which it grew larger and became painful. The mass was excised elsewhere, in 1916, at which time it was 6 cm. in diameter. There is no pathologic note on the specimen. One year later, in 1917, she noted a recurrence in the upper outer portion of the scar of the incision. This has gradually increased in size during the past three years and during the past two weeks it has become painful. Examination showed a normal thyroid gland. The left breast was enlarged and showed a scar 14 cm. long at the site of the previous excision. In the midportion of the scar there were two masses, one, 9 cm. in diameter internally to the scar and another 21.5 cm. in diameter lateral to the scar. The overlying skin is attached to the masses and is slightly edematous. Both masses gave the sensation of fluid under tension on palpation, and over the larger mass there is an area 4 cm. in diameter, which gives the impression of fluctuation. Two enlarged lymph nodes can be palpated in the left axilla. A radical operation was performed July 28, 1920. Both the masses showed hemorrhagic and grey, glistening, translucent material on section. In the larger tumor a small cyst was found. Pathologic examination showed definite gelatinous carcinoma. There are no metastases to nodes, the large axillary nodes showing tuberculous adenitis. The patient reported that she was well in 1924, was not heard from again. Her attending physician reports that she died of recurrent car-

cinoma of the breast February 24, 1938, 18 years after the radical operation and 22 years after the first excision.

**Path. No. 17869.**—The patient, at the age of 36, in 1896, had her left breast removed for cancer. There is no note on the duration of symptoms or clinical findings prior to operation. The pectoralis minor was not removed and part of the axilla was not dissected. A pathologic note by Dr. G. P. Briggs of New York, in 1896, states

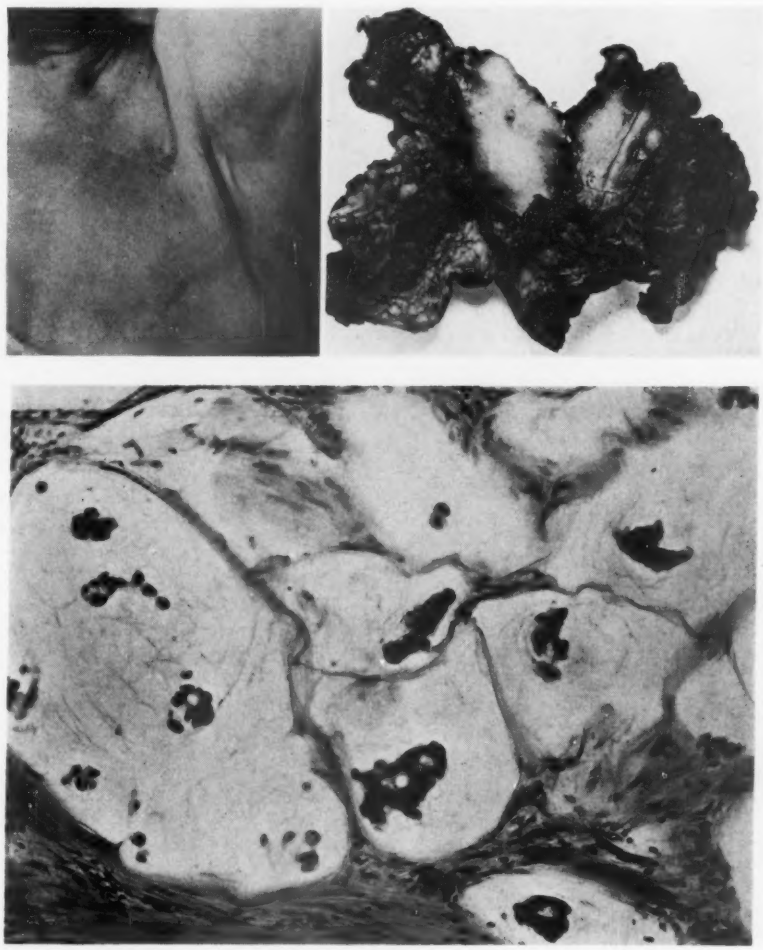



FIG. 20A, B and C.—Path. No. 17869: Photograph of the patient, gross specimen and photomicrograph of a case of gelatinous carcinoma treated by mastectomy, in 1896. Dissection of the axilla and removal of the pectoral muscle with recurrent cancer was performed, in 1915. The patient died, in 1931, with mediastinal metastases, 35 years after the first operation.

that the tissue removed last February, consists chiefly of development of myxomatous tissue in the walls of the alveoli, the tumor removed is unquestionably a carcinoma which has undergone early degeneration into a mucoid substance of indefinite character. The degeneration has involved chiefly the carcinomatous cells, beginning in those nearest the alveolar wall and often leaving a central bunch of cells which are nearly normal.

Where breast tissue can still be recognized there is often seen proliferation of epithelial cells and infiltration of the fibrous stroma with small round cells. Tumors presenting extensive degeneration into homogeneous gelatinous material have usually been designated as colloid and I think this breast tumor is most properly called a "colloid carcinoma."

This letter of Dr. Briggs, written in 1896, describes the tumor removed by Dr. Bull.

Nine years later a mass appeared just below the axilla which gradually increased in size. In July, 1915, Dr. Bloodgood operated upon the recurrent tumor and performed a partial excision of the mass in the apex of the axilla. He found it impossible to remove all the tumor because of its proximity of the larger nerves and vessels. Following this, the patient remained fairly well, but, in 1919, a mass appeared in the region of the clavicle. The patient received deep roentgenotherapy, in 1919, 1920, and in 1922. In 1926, an indurated mass developed, suggesting a recurrence beneath the clavicle. Further irradiation was given. In April, 1931, Dr. Dandy performed an operation for the relief of pain because of symptoms of nerve pressure in the brachial plexus. At this operation tumor tissue, microscopically verified, was removed from the nerves. Pain, however, soon recurred with swelling of the arm and lymphedema.

In November, 1931, the arm was amputated. Erysipelas developed in the wound and the patient died, November 21, 1931, 35 years after the first operation with mediastinal metastases. At all the operations, subsequent to 1896, mucoid carcinoma could be demonstrated in the axilla or supraclavicular regions (Fig. 20 A, B and C).

In diffuse gelatinous carcinoma with malignant epithelial cells of the scirrhus type, 10 cases had metastatic involvement of the lymph nodes. Only one of these patients was cured and this one remained well 14 years. Two died five years later of the disease and another had metastases to the opposite breast at the end of five years. One was dead four years later of other causes.

Six cases of gelatinous carcinoma with basal cells had metastases to the axillary lymph nodes. One died postoperatively, and another one year later of accidental death. Three died of the disease, 7, 18 and 35 years, respectively, after radical operation, and one was reported well 16 years later.

Twenty-four cases in the present series showed partial mucoid change. The predominant histologic picture, with few exceptions, was that of scirrhus carcinoma. The exceptional cases were comedo cancer (duct cancer) with areas of gelatinous carcinoma. Twenty of these cases have been adequately traced. Six survived the five-year period, and 14 are known to be dead of the disease. Eight of these cases showed skin involvement at the time of the first examination and 16 out of 24 had axillary involvement. On the other hand, in instances of gelatinous carcinoma with diffuse mucoid change, metastases occurred in 19 out of 57 cases, or approximately one-third. In the fatal cases the lungs were the most frequent site for internal metastases. Such involvement was reported in six cases. The liver, brain and skeleton were each involved twice.

## CONCLUSIONS

Gelatinous carcinoma is a rare form of mammary cancer, and occurred 83 times in a series of 2,300 breast cancers. The peak of age incidence is between 46 and 50 years compared to 41 and 45 years for scirrhus cancer. The

TABLE III

SYNOPSIS OF DATA RELEVANT TO 59 CASES OF GELATINOUS CARCINOMA

| Path. No. | Age | Duration | Clinical | Operation | | Result |
|---|---|---|---|---|---|---|
| 60479 | 62-C | 2 yrs. | 6 cm., near nipple; nipple bulges; skin involvement | C.C.* | 5-22-35 | Dead June, 1937 |
| 58398 | 55 | 3 mos. | Multiple nodules; skin red | Excision | 8- 7-37 | |
| 58216 | 34 | 21 mos. | | C.C. right | 1-15-34 | Well 4-21-38 |
| | | | | C.C. left | 6-21-37 | |
| 57602 | 67 | 4 mos. | 4 cm., L.O.Q.; black and blue cyst | Excision | 2- 3-37 | |
| | | | | C.C. | 2- 5-37 | |
| 57290 | 41 | 4 mos. | 3 cm. Clinically cyst gel flowed from wound | Excision | 11-18-36 | |
| | | | | C.C. | 11-20-36 | |
| 56134 | 46 | 1 yr. | 3 cm., central | C.C. | 2-10-35 | Well Sept., 1937 |
| 55693 | 68 | 4 yrs. | 10 cm., near nipple; 3 lumps felt; skin involvement | C.C. | 9-25-33 | Well 2-22-38 |
| 55022 | 69 | 6 mos. | 5 cm., U.I.Q.; clinically cyst | Excision | 3- 5-35 | Well 3-28-38 |
| 52578 | 52 | 1 yr. | 3 cm., U.O.Q.; nipple protrudes | C.C. | 1- 6-34 | Dead 3-19-38 |
| 52081 | 76 | 2 wks. | 3 cm., above nipple; second nodule above; nipple retracted | C.C. | 9- 6-32 | Dead 7-7-36, other causes |
| 51228 | 50 | 6 mos. | 3 cm., beneath nipple | C.C. right | 5-15-33 | Metastases left axilla 2-24-38 |
| | | | | Excision left | 1-10-38 | |
| 50507 | 48 | 14 yrs. | 5 cm.; skin discolored | Excision papilloma | 1902 | Well 2-27-38 |
| | | | | C.C. | 1925 | |
| | | | | Excision | 11-10-31 | |
| | | | | Excision | 10-18-32 | |
| 49620 | 52 | 3 yrs. | 8 cm. | Simple mastectomy | 12- 6-32 | Well 2-27-38 |
| 48052 | 40 | | 4 cm., near nipple | C.C. | 5-10-32 | Well 3-7-38 |
| 47241 | 45 | 3 mos. | Near nipple | C.C. | 4-15-30 | Well 2-28-38 |
| 45703 | 44-C | Found routine | 3 cm., U.O.Q. | Excision | 8- 5-29 | Lost |
| | | | | C.C. | 8-10-29 | |
| 44868 | 41 | 2 mos. | Central; nipple discharge | C.C. | 5-10-31 | Well 1-15-38 |
| 43462 | 66 | 3 days | 2 cm., U.I.Q. | Excision | Aug., 1930 | Well 2-22-38 |
| | | | | C.C. | Aug., 1930 | |
| 41940 | 34 | 9 mos. | 2 cm., central | Excision | 7-23-29 | Dead 4-26-35 |
| | | | | C.C. | 7-24-29 | |
| 41018 | | | | Excision | 10-29-30 | |
| 39396 | 57 | 3 mos. | 8 cm., L.O.Q.; skin red | C.C. | 4- 4-27 | Well 2-26-38 |
| 39095 | 46 | | 5 cm. | | | |
| 38598 | 79 | 48 yrs. | 5 cm., U.I.Q.; nipple bulges | C.C. | 12- 7-26 | Dead 8-6-35, old age |
| 37682 | 40 | 2 wks. | 6 cm., near nipple; lump in other breast | C.C. | 5-24-23 | Dead 9-15-28 |
| 36862 | 55 | 9 yrs. | 2 cm.; clinically cyst | C.C. | 7-16-25 | |
| 36257 | 36-C | 6 yrs. | Entire breast | C.C. | 10-17-24 | Dead 2-11-31 |
| 35405 | 44 | 2 yrs. | 0.5 cm.; tumor previously removed opp. breast, U.O.Q. | C.C. | 5-13-24 | Dead 2-17-25, other causes |
| 35154 | 44 | | 2 cm., upper brea t | Excision | April, 1924 | Well 5-12-30 |
| | | | | X-ray therapy p.o. | | |
| 34353 | 66 | 8 yrs. | 2 cm., central; bleeding and protrusion of nipple | C.C. | 12- 8-23 | Well 5-17-30 |
| 34033 | 48 | 3 yrs. | 6 cm., U.O.Q.; clinically cyst | C.C. | 10- 6-23 | Well 3-14-38 |
| 32651 | 40 | 3 mos. | 2 cm., near nipple; breast cystic | C.C. | 4- 6-23 | Metastases 1937 |
| 30464 | 55 | 1 yr. | 6 cm., central; skin discolored | C.C. | 5-29-22 | Dead 2-12-25 |
| 30448 | 60 | | | C.C. | 1920 | Dead 1924, heart disease |
| 29851 | | | Autopsy specimen | Explored | 1922 | Dead Dec., 1924, metastases to bone |

*C.C.=Complete operation for cancer.

SYNOPSIS OF DATA RELEVANT TO 59 CASES OF GELATINOUS CARCINOMA—*Continued*

| Path. No. | Age | Duration | Clinical | Operation | | Result |
|---|---|---|---|---|---|---|
| 29697 | 32 | 2 yrs. | 2 cm.; clinically cyst | Excision | 10-29-19 | Well 4-6-38 |
| | | | | Mastectomy | Feb., 1922 | |
| | | | | Excision axillary node | Nov., 1930 | |
| 27946 | 50 | | | C.C. | 1921 | Well 3-30-35 |
| 27564 | 37 | 13 mos. | Two lumps | C.C. | 2- 7-21 | Dead 4-10-22, brain metastases |
| 26200 | 40 | 6 yrs. | 10 cm. | C.C. | 6- 3-20 | Dead 2-24-38, recurrent cancer |
| 25887 | 38 | 4 mos. | Bleeding from nipple | C.C. | 10-16-19 | Dead 1921, metastases to lung |
| 25249 | 30 | 5 mos. | | C.C. left | 10-29-19 | Dead 1-29-21 |
| | | | | C.C. right | 11- 2-20 | |
| 24495 | 94 | | Autopsy specimen | None | | |
| 24433 | 70 | 2 yrs. | 8 cm., U.O.Q. | C.C. | 4-28-19 | Dead P.O. |
| 24144 | 43 | 2½ yrs. | 4 cm., central; clinically fibroadenoma | C.C. | 2-27-19 | Well 1930 |
| 21918 | 68 | 2½ yrs. | Bleeding from nipple | Mastectomy | 7-26-17 | Dead 1919, heart disease |
| 18441 | 49 | 3 mos. | | Mastectomy | 11- 6-15 | Well 1931 |
| 17869 | 36 | | Recurrence after 20 years | Mastectomy | 1896 | Dead 11-21-31, cancer |
| | | | | C.C. | 7-22-15 | |
| | | | | Irradiation | 1919-1922 | |
| 17378 | 64 | 20 yrs. | 3 cm.; discharge nipple; breast indurated | C.C. | 4-17-15 | Dead 1921 |
| 10011 | 43 | 6 yrs. | 4 cm., U.O.Q.; clinically fibroadenoma | C.C. | 10- 6-09 | Lost |
| 9260 | 58 | 2 mos. | 5 cm., U.I.Q. | C.C. | 10- 1-08 | Dead 1909 |
| 8209 | 46 | 2 wks. | 2 cm., U.O.Q. | C.C. | 5-16-07 | Dead May, 1912 |
| 7101 | 69 | 3 mos. | 5 cm., above nipple | C.C. | 2-22-06 | Well Aug., 1919 |
| 6821 | 40 | 1 mo. | 2 cm., U.I.Q.; clinically cyst; bleeding from nipple | C.C. | 11-10-05 | Well 1924 |
| 5278 | 46 | 6 mos. | 5 cm., O.U.Q.; skin involvement | C.C. | 2-11-04 | Dead 1905 |
| 4919 | 52 | | Central; bleeding from nipple | C.C. | 6-23-99 | Dead July, 1902 |
| 4918 | 51 | | Beneath nipple | Mastectomy | 6- 7-02 | Lost |
| 4874 | 43 | 9 mos. | 5 cm., beneath nipple; bleeding from nipple | C.C. | 6- 4-03 | Well 1930 |
| 4786 | 49 | 4 yrs. | 4 cm. | Two previous excisions | 1895-1899 | Dead 1931, accident |
| | | | | Excision | 4-23-03 | |
| | | | | Excision | 6- 6-06 | |
| 4204 | 45 | 4 yrs. | 7 cm., beneath nipple | C.C. | 2- 4-00 | Dead 1-2-04 |
| | | | | Excision nodes neck | 4-15-02 | |
| 795 | 58 | 6 mos. | 3 cm., I.U.Q.; skin involvement | C.C. | 3- 2-95 | Dead Nov., 1904, pneumonia |

discovery of a lump was the first sign noted by the patient in 80 per cent of the cases and in nearly half the cases the known duration of the mass prior to examination was a year or more, averaging 4.3 years. The average diameter of the tumor in the present series was 4.6 cm. and only four tumors were 10 cm. or over in diameter. The most prevalent sites in the breast are the outer upper quadrant and central zone.

There are four chief findings on clinical examination which suggest a

diagnosis of gelatinous carcinoma: The first, is the relatively small size of the tumor in comparison with the long duration of symptoms. The second, is the protrusion and enlargement of the nipple on the affected side. The third, is the cystic character of the growth on palpation, the growth being differentiated from a benign cyst by aspiration of characteristic mucoid material. The fourth, is the presence of a "swish" on firm pressure, as originally described by Halsted.

TABLE IV

SYNOPSIS OF DATA RELEVANT TO 24 CASES OF SCIRRHUS CANCER WITH MUCOID CHANGE

| Path. No. | Age | Duration | Clinical | Operation | | Result |
|---|---|---|---|---|---|---|
| 48446 | | | | C.C. | 7- 4-32 | Dead 3-15-33, metastases |
| 45703 | 44-C | Routine examination | 3 cm., upper outer quadrant | Excision C.C. | 8- 5-29 | Lost |
| 41029 | 28 | 1 yr. | | C.C. | 3-26-27 | Dead 7-26-28, heart disease |
| 41018 | | | | Excision | 10-29-30 | Lost |
| 40882 | 48 | 4 yrs. | 0.5 cm.; recurrent | C.C. Excision twice | 10- 9-28 | Dead 1930, metastases to spine |
| 39347 | 73 | | 5 cm.; cystic | Excision | 5-29-26 | Well 1932 |
| 39146 | 68 | 18 mos. | Discharge nipple; ulceration | C.C. | 3-16-27 | Dead Sept., 1933, phlebitis |
| 39112 | 47 | 18 mos. | Large tumor | C.C. | 4-14-27 | Dead 1928, metastases to lungs |
| 38540 | 34 | 3 wks. | 3 cm., nipple zone | Excision C.C. | 11-12-26 11-18-26 | Dead 11-23-27, metastases to brain |
| 37408 | 40 | 4 mos. | Diffuse; two tumors | C.C. | 12-26-25 | Dead June, 1934, cancer |
| 32416 | | 1 yr. | 5 cm.; nipple freely movable | C.C. | 1-31-23 | Dead 1926, metastases to lung |
| 31853 | | | Cystic | Excision | 11-28-22 | Dead |
| 26068 | 32 | 2 wks. | | C.C. | 5- 2-20 | Dead 2-7-24, carcinoma stomach |
| 23633 | 54 | 11 mos. | 6 cm., near nipple | Excision C.C. | 12- 6-15 8-27-18 | Well 3-4-38 |
| 18592 | 78 | 6 mos. | | | | Dead 1919, metastases |
| 16076 | 39 | 2 yrs. | 1 cm.; skin changes; breast infiltrated | C.C. | 7-24-14 | Dead 8-6-15, metastases to lung |
| 15549 | 43 | 2 days. | | C.C. | 4- 1-17 | Well 2-23-25 Lost 1927 |
| 10630 | 48 | 5 yrs. | 14 cm., upper hemisphere | C.C. | 6- 6-10 | Dead 1911 |
| 9733 | 32 | 2 yrs. | 8 cm., nipple zone; pig-skin | C.C. | 6- 4-09 | Dead 1914 |
| 9077 | 37 | 3 mos. | Outer lower quadrant; skin nodules | C.C. | 7- 2-08 | Dead 1909 |
| 8467 | 46 | 9 mos. | | C.C. | 9-16-07 | Dead 1909 |
| 8039 | 31 | 8 mos. | | C.C. | 3-21-07 | Dead 1908 |
| 7567 | 49 | 3 yrs. | 9 cm., nipple zone; nodes palpable | C.C. | 9-21-06 | Lost |
| 5078 | 45 | 2 mos. | 10 cm.; skin metastases; nodes palpable | C.C. | 1904 | Dead 3 mos. later |

Mucoid cancers on gross and microscopic examination may be diffusely or partially gelatinous. The gelatinous material has a characteristic gray, translucent appearance, resembling tapioca. Microscopically, mammary can-

cers with diffuse gelatinous change originate in papillary cancer, adenocystic basal cell cancer, or slowly growing adenocarcinoma. The mucoid material is secreted by the epithelial cells of the tumor. Cancers showing partial mucoid change are usually of the scirrhus type. In the present series, 59 of 83 cases showed diffuse or typical gelatinous change. Thirty-four of 45 of these patients, who were adequately traced, survived the five-year period—75 per cent of five-year cures. Six of these patients eventually died of recurrent disease, one case 16 years, another 18 years after radical mastectomy.

Twenty of 24 cases with carcinomata showing partial mucoid change were adequately traced. Only five survived the five-year period—30 per cent of five-year cures. In the present series, the five-year survivals for all types of mammary carcinomata showing mucoid change (diffuse or partial) was 60 per cent.

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# THE DIFFERENTIAL DIAGNOSIS OF HYPERPARATHYROIDISM*

## WITH SPECIAL REFERENCE TO POLYOSTOTIC FIBROUS DYSPLASIA

## (*Lichtenstein—Jaffe*)

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IN THE 12 years that have passed since Mandl, at the suggestion of Erdheim, first removed a parathyroid adenoma in a case of hyperparathyroidism, the number of reported cases of this disease has increased considerably. Up until February, 1936, Wilder and Howell were able to collect 135 cases, which, upon careful analysis, were unquestionably authentic instances of the disease. Undoubtedly, there have been many others which have not been reported.

The clinical, roentgenographic, and chemical aspects of hyperparathyroidism have been stressed so frequently in the past decade that the disease has become familiar to the medical profession at large.

It may be well, however, to again state that the various manifestations of hyperparathyroidism are dependent upon the secretory hyperactivity of one or more parathyroid adenomata which bring about a profound disturbance of calcium and phosphorus metabolism, and that surgical removal of the tumor results in either complete cure or marked amelioration of the symptoms. The disease, which occurs more frequently in females and usually in middle life, is measured, as a rule, in terms of years. It is characterized by bone and joint pain, muscle weakness, localized bone swellings, pathologic fractures, particularly of the extremities and ribs, disturbances of gait, and, in advanced cases, deformities of the bones. There may be other symptoms which become so prominent as to cloud the more important aspects of the clinical picture. These are attacks of intractable nausea and vomiting, polyuria and polydypsia, renal colic, anorexia, severe constipation, loss of weight, and secondary anemia.

The explanation of the roentgenologic findings rests upon a knowledge of the disturbance of physiologic activity of the parathyroid tumor. Normally, it is the function of the parathyroid bodies to control calcium and phosphorus metabolism within the narrow confines of fairly constant blood serum values of 9.5 to 10.5 mg. of calcium per 100 cc. and 3 to 3.5 mg. of phosphorus per 100 cc. When parathyroid activity is increased, because of the presence of a hyperfunctioning tumor, the serum calcium level is increased and the phosphorus decreased due to the fact that greater quantities of calcium salts are withdrawn from the bones. Usually, increased phosphatase activity can be demonstrated. The effect of prolonged withdrawal of calcium salts from the skeleton becomes evident upon roentgenologic examination. The bones of the

* Read before the New York Surgical Society, January 26, 1938. Submitted for publication December 28, 1937.

skull present a finely granular appearance. The long bones appear porotic with thinning of the cortex and trabeculae. There may be cyst formation in the center of the shaft. The pelvic bones are frequently cystic. The vertebrae present a coarsely granular pattern similar to that seen in the skull. Due to softening of the skeleton, deformities result from gradual collapse of supporting structures such as the spine, pelvis and thoracic cage.

Up to the present time, most observers have agreed that a diagnosis of hyperparathyroidism should be made when, in addition to the clinical symptoms and roentgenologic findings already enumerated, there is found a hypercalcemia, a hypophosphatemia, an increase in the serum phosphatase and a proven negative calcium balance. In fact, the combination of these laboratory findings is considered pathognomonic of the disease. However, the course of events and the laboratory findings in Case 3, herewith appended, tend to cast considerable doubt as to the validity of considering these laboratory tests as pathognomonic of hyperparathyroidism. As a result of this experience it may become necessary to reconsider the entire problem in order to establish criteria which would aid in the increasingly difficult aspects of differential diagnosis.

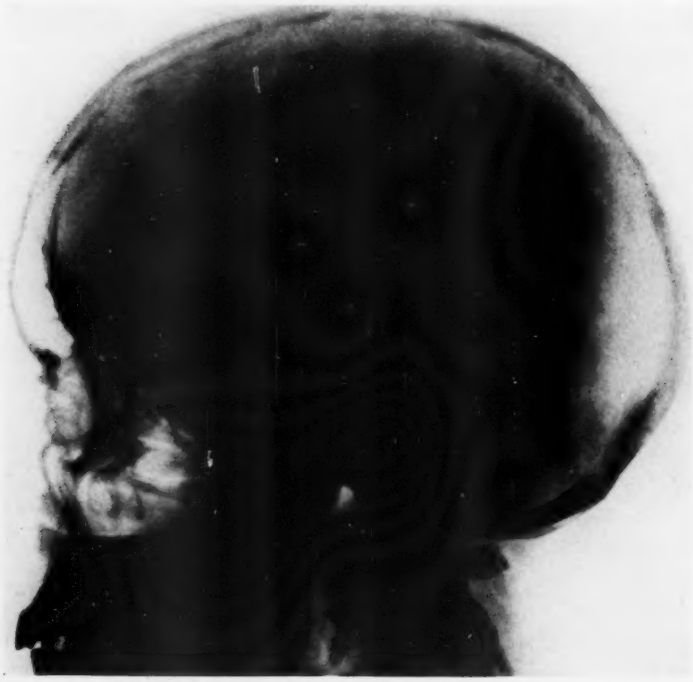

FIG. 1.—Case 1: Roentgenogram of skull showing a finely granular appearance of the bones of the calvarium due to absorption of calcium.

In order to emphasize some of the points to be considered in differential diagnosis, two cases of proven hyperparathyroidism are briefly reported.

**Case 1.**—M. L., female, age 29, single, referred by Dr. Reuben Ottenberg on March 30, 1937. About 18 months previously, she had consulted her dentist concerning the re-

moval of a tooth. Roentgenologic examination of the jaw at the time showed bony rarefaction involving the maxilla and mandible. A few months later, after a comparatively slight injury, the patient fractured the left patella, which, however, united without much difficulty. About one year ago, roentgenologic examination of the long bones demonstrated small cystic areas in the lower ends of the right radius and ulna. At that time the blood calcium was reported to be 14 mg. per 100 cc., and the phosphorus 2 mg.

A roentgenologic examination of the entire skeleton showed: (a) Marked decalcification of the skull, which presented a sieve-like appearance; (b) the radius and ulna were decalcified, with small cystic areas in the proximal end of the left radius and distal ends of right radius and ulna; (c) the knees showed marked bony absorption in the femora, with cysts in the left patella; (d) the pelvis showed cystic bone absorption in the right ilium and a suspicious area in the left sacrum. Roentgenologic examination of the kidneys showed no calculi (Figs. 1, 2 and 3).

Physical examination was negative. Urine and blood examinations showed no abnormalities. Just previous to admission to the hospital, the serum calcium was 11.8 mg., phosphorus 2.4 mg. and phosphatase 30.5 Bodansky units.

The patient was admitted to the Mt. Sinai Hospital April 7, 1937. Calcium balance studies showed that, after a three-day diet containing 300 mg. of calcium, the excre-

FIG. 2.—Case 1: Roentgenogram of left knee showing cyst in left patella and coarse trabeculation of the femur and tibia.

FIG. 3.—Case 1: Roentgenogram of pelvis showing cystic disease involving the ilium.

tion of calcium was 1,073 mg., 771 mg. appearing in the urine and the remainder in the feces. In other words, there was a negative calcium balance of 773 mg.

*Operation.*—April 13, 1937: A parathyroid adenoma measuring 4.5 by 2 by 1 cm. was found at the lower pole of the right lobe of the thyroid gland (Fig. 4). It dipped downward, behind the sternum, toward the mediastinum. Both sides of the neck were explored, but no other tumors were found. Two normal parathyroid bodies were found on the left side.

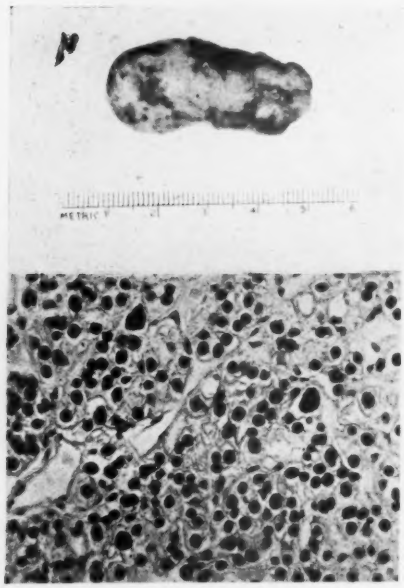

FIG. 4.—Case 1: Photograph of tumor of the parathyroid removed at operation. Microscopic section of it which shows that the tumor was composed mainly of chief cells.

*Pathologic Examination* of the tumor showed it to be quite cellular. It was composed mainly of pale, water-clear chief cells, with frequent giant cells, and some small groups of oxyphile cells (Fig. 4).

Convalescence was uneventful until the fourth day, when she developed headache, tingling in the face, hands and feet, and diarrhea. There was a positive Chvostek's sign. At this time the serum calcium was 6.9 mg. and the phosphorus 3 mg. This mild tetany responded promptly to parathormone and calcium gluconate. At the time she left the hospital, April 23, 1937, the serum calcium was 9 mg.

Since her discharge, the patient has improved steadily. There has been no recurrence of symptoms and the blood figures are normal. The patient has returned to work.

**Case 2.**—Hosp. No. 407735: An Italian woman, age 36, was admitted to the Medical Service of the Mt. Sinai Hospital September 8, 1936. For the preceding year she had been receiving treatment in the Out-Patient Department for a right renal calculus. She complained of recurring attacks of pain in the right lumbar region, which had begun four years previously, following a pregnancy. The pain occasionally radiated to the right groin. It was never accompanied by chills, fever or hematuria. The past history was irrelevant.

Physical examination was negative. The blood count was normal, as was also the urinalysis. Blood sugar, 90 mg.; urea nitrogen, 15 mg.; serum calcium, 13 mg.; serum phosphorus, 4 mg. Phosphatase determination showed eight King-Armstrong units. A later blood examination showed the calcium to be 11.8 mg. and the phosphorus 3.5 mg. Roentgenologic examination of the skull and long bones failed to reveal any abnormality. A suspicious rarefied cystic-like area was seen in the left ilium. The patient was discharged September 30, 1936, as a possible case of hyperparathyroidism.

She was readmitted April 26, 1937, complaining that she had been having generalized bone pains for the preceding three months. These were boring in character and involved mainly the left shoulder, knees and hips. She also experienced occasional attacks of nausea without vomiting.

Physical examination was again negative. The roentgenologic examination was repeated and disclosed an enlargement of the previously noted cystic area in the left ilium, as well as the presence of two small calculi in the right kidney pelvis (Fig. 5). Serum calcium, 11.6 mg.; phosphorus, 3.4 mg.; calcium balance studies revealed a daily negative balance of one gram.

FIG. 5.—Case 2: Roentgenogram of pelvis showing cystic rarefaction in the left ilium.

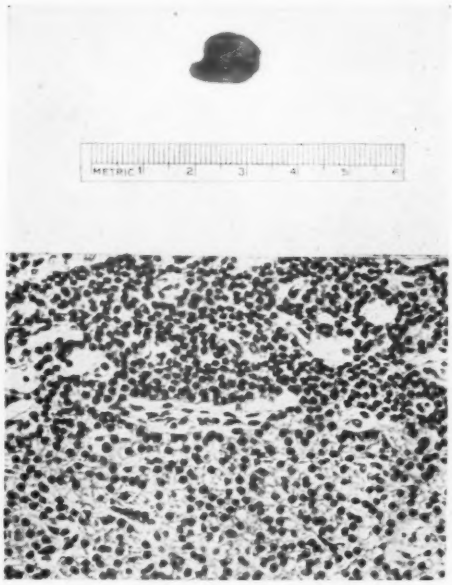

FIG. 6.—Case 2: Photograph of tumor of a parathyroid removed at operation. This represents about two-thirds of the original size of the tumor. Microscopic section of it shows that the tumor was composed mainly of chief cells.

FIG. 7.—Case 3: Roentgenogram of the left hip showing an incomplete fracture of the neck of the femur and the apparent cystic rarefaction of the neck and shaft of the femur.

FIG. 8.—Case 3: Roentgenogram of the skull showing the finely granular appearance similar to that seen in Fig. 1. In addition, there is apparent cystic rarefaction of the occipital protuberance.

*Operation.*—April 30, 1937: Considerable difficulty was encountered in locating the parathyroid adenoma, which was finally found embedded in the substance of the right lobe of the thyroid near its postero-external surface. The adenoma which measured 3 by 2.5 by 1.5 cm., presented the typical reddish-brown color. It was removed *in toto*. Search was made for additional tumors, but none were found. Two normal parathyroid bodies were demonstrated. Histologic examination of the adenoma showed it to be composed mainly of chief cells (Fig. 6).

Convalescence was uneventful other than for the development of a positive Chvostek's sign which persisted for one week. The serum calcium figures were 8.7, 9.7 and 9.1 mg. The patient was discharged May 15 in excellent condition.

When last seen, November 8, 1937, she stated that she no longer had any bone pains and that the pain in the right lumbar region had also disappeared.

FIG. 9.—Case 3: Roentgenogram of the right humerus which presents an appearance similar to that seen in osteitis fibrosa cystica. Closer examination indicates thinning of the cortex which, on microscopic section, is found to be due to erosion from the endosteal surface. The apparent cystic areas represent fibrous tissue containing islands of immature bone.

In contradistinction to the above cases, I wish to report, in detail, the history of the patient which forms the basis of this paper.

**Case 3.**—M. S., male, age 47, was seen May 2, 1937 in Richmond, Va., in consultation with Drs. Wm. H. Higgins, Carrington Williams, Bigger and Alice Bernheim. Approximately two months previously, while in England, the patient had developed pain and lameness in the left hip. Three weeks later while playing ping-pong on board ship, he experienced a sudden, sharp pain in the left hip. He was able, however, to carry on his activities, but with some difficulty. Roentgenologic examination demonstrated an incomplete fracture of the neck of the left femur without displacement or impaction (Fig. 7). In addition, there was found an extensive rarefaction of the neck and shaft of the femur

which suggested to Doctor Higgins the possibility of osteitis fibrosa cystica. He was admitted to St. Luke's Hospital, Richmond, for study.

Roentgenologic examination of the remainder of the skeleton disclosed what appeared to be cystic rarefactions in the lower end of the left femur, lower end of right humerus, occipital bone, pelvis and two ribs (Figs. 8, 9 and 10). Blood serum examination showed calcium, 13 mg.; and phosphorus, 2.8 mg. per 100 cc.

On the natural assumption that the patient was suffering from hyperparathyroidism resulting in a pathologic fracture of the left hip, exploration of the neck was undertaken by Dr. Carrington Williams. After a prolonged and thorough search, no parathyroid adenoma was demonstrable. Convalescence was uneventful and the wound healed by first intention.

FIG. 10.—Case 3: Roentgenogram of left femur. The findings here are similar to those seen in Fig. 9. They may be easily mistaken for the roentgenologic evidence one sees in cases of osteitis fibrosa cystica.

Careful inquiry into the patient's past history brought out the interesting fact that since childhood the left leg had been one inch longer than the right, and that for the past ten years the patient had had frequent pains in the left thigh and knee.

The patient was transferred to New York in order that careful calcium balance studies might be undertaken in the hope of definitely establishing a positive diagnosis, and entered the New York Hospital May 4, 1937.

Examination was negative except for a prominent occipital protuberance and the fact that the left lower extremity was 3 cm. longer than the right.

*Laboratory Data.*—Urine, negative; there was no Bence-Jones protein; hemoglobin, 94 per cent; red blood cells, 4,570,000; white blood cells, 8,600; polymorphonuclear leukocytes, 66 per cent; lymphocytes, 19 per cent; monocytes, 11 per cent; basophiles, 4 per cent; serum calcium, 11.3 mg.; phosphorus, 3.2 mg.; and phosphatase, four Bodansky units.

The patient was placed on a three-day diet containing 300 mg. of calcium. During this time, bowel function was normal. The diet was continued for three more days and all urine and feces excreted during this second period were examined for calcium excretion. The measured calcium intake during the initial three-day period obviates the possibility of error from additional calcium which may be present in the intestinal tract. Examination of the stool and urine specimens collected during the second period showed an excretion of 1547 mg. of calcium, 841 mg. appearing in the urine and 706 mg. in the feces. In other words, the patient presented a marked negative calcium balance. This seemed fairly conclusive confirmatory evidence of the existence of hyperparathyroidism.

Before undertaking a second neck exploration, I asked Dr. H. Jaffee to see the patient. After a prolonged examination of the roentgenograms and the laboratory data, he formed the opinion that the evidence was overwhelmingly in favor of a diagnosis of hyperparathyroidism, but reserved a 5 per cent possibility that the patient might be suffering from polyostotic fibrous dysplasia. He concurred in the opinion that the neck should be explored again.

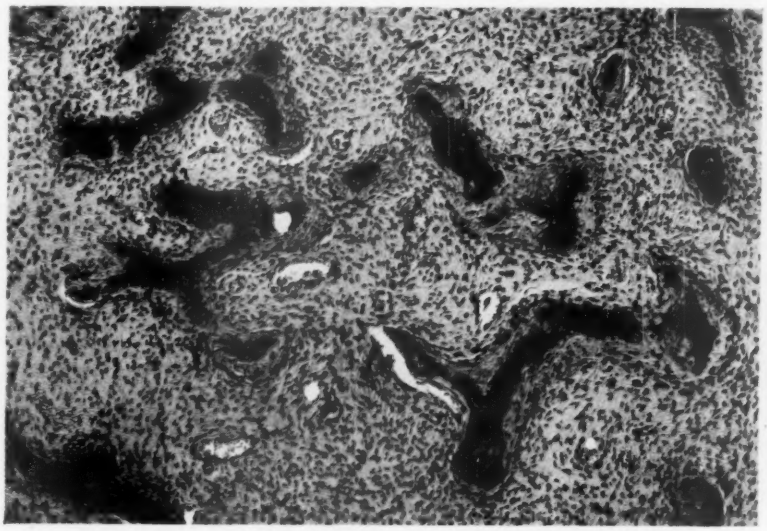

FIG. 11.—Case 3: Microscopic section of the bone biopsy taken from the left femur which shows fibrous tissue metaplasia of the bone marrow containing scattered islands of immature bone. This is one of the characteristic findings of "polyostotic fibrous dysplasia."

*Operation.*—May 14, 1937: The technical difficulties were considerable because of extensive scarring. A thorough exploration of the neck and superior mediastinum was made, but no parathyroid adenoma was found. The wound was closed. Through a longitudinal incision on the lateral aspect of the left thigh, the upper part of the femur was exposed and a liberal section of cortex and subjacent tissue was removed. The cortex was found to be extensively eburnated and somewhat thinned. The marrow was replaced by dense fibrous tissue which had a rubbery consistency.

*Pathologic Examination* of the bone biopsy material by Doctor Jaffe showed replacement of the normal marrow by vascular fibrous tissue which contained numerous trabeculae of immature bone (Fig. 11). There were no giant cells and no evidences of active resorption or transformation of the metaplastic new bone. *Diagnosis.*—Polyostotic fibrous dysplasia.

Convalescence was uneventful. The operative wound healed without incident and the patient returned to Richmond the end of May. Repeated roentgenologic examinations of the hip showed subsequent solid union at the site of fracture. At Doctor Bernheim's sug-

gestion the patient was placed on a high calcium and high vitamin diet which he has continued to date. He was advised to avoid undue physical activity in order to guard against possible fracture. He has resumed his business activity and now feels well. A recent check-up roentgenologic examination of the skeleton indicates that some calcification of the involved bones is taking place. The serum calcium is 8.7 mg.; phosphorus, 3.4 mg.; phosphatase, 5.9 Bodansky units.

Discussion.—There will appear in the Archives of Surgery, vol. 36, 874–898, May, 1938, an article by L. Lichtenstein on "Polyostotic Fibrous Dysplasia." The material forming the basis of this paper was assembled in the laboratory of Dr. H. Jaffe at the Hospital for Joint Diseases in New York. The following discussion of the disease is based upon conversations with Drs. Jaffe and Lichtenstein, to whom appreciation is hereby expressed for the privilege of reporting, briefly, the results of their investigations.

The disease, which Lichtenstein gives the name "Polyostotic Fibrous Dysplasia," has been, heretofore, reported in the literature under a great variety of titles, such as "Osteo-Dystrophia Fibrosa Unilateralis," "Unilateral Recklinghausen's Disease," "Unilateral Polyostotic Osteitis Fibrosa," "Focal Osteitis Fibrosa," "Osteitis Fibrosa in Multiple Foci," "Osteitis Fibrosa with Formation of Hyaline Cartilage," "Osteitis Fibrosa Disseminata," *etc.* Lichtenstein and Jaffe have, up to the present time, seen nine instances of the disease, four in their own hospital and five at other institutions, including the case reported in this paper.

A review of the case histories would seem to indicate that the age of onset of symptoms is in childhood or early adolescence, and that females are predominantly affected. The common presenting symptoms are limp, bone pain, deformity of the affected limb and pathologic fracture. It may take years for the disease to progress to a point where medical aid is sought. Our patient was age 47 before severe symptoms developed, although bone pain had been present for the preceding 10 or 12 years.

A peculiar feature is the tendency of the bone lesions to be predominantly unilateral in distribution, either side being affected without preference. Exceptions to this, however, are not uncommon, as is indicated in our own case. Although the femur and tibia are most frequently involved, it is not unusual to find evidences of the disease in the radius and humerus as well. The skull and pelvis may also be affected.

The characteristic findings on roentgenologic examination may be summarized as follows: (1) Broadening or expansion of the bone; (2) thinning of the cortex; (3) characteristic rarefied and apparently trabeculated appearance; (4) secondary deformities of the affected bones. Pathologic fracture of the neck of the femur is common. Inasmuch as the condition is frequently erroneously interpreted as osteitis fibrosa cystica, or Recklinghausen's disease, it is important to examine the entire skeleton roentgenologically in order to determine whether the bone lesions are unilateral in distribution. This is an important point in differential diagnosis. As will be pointed out, the bones involved in polyostotic fibrous dysplasia do not contain cysts, in contradistinc-

tion to hyperparathyroidism, and this must be borne in mind when interpreting the roentgenograms.

The basis of Lichtenstein's interpretation of the pathologic features of the disease is adequate biopsy material obtained from nine cases. The bone cortex is considerably thinned out, due in part to resorption, but largely to erosion of the endosteal surface by the proliferating fibrous tissue replacing the marrow cavity. There is no evidence of periosteal proliferation or new bone deposition. The medullary cavity is filled with fibrous tissue which is grayish-white in color and has a peculiar consistency described as spongy or rubbery. It has also been noted to be gritty. This fibrous tissue is composed of spindle cells with oval, pale staining nuclei. In some areas the basic connective tissue has undergone a fibroblastic differentiation into mature connective tissue containing a large amount of collagen. Dispersed irregularly in this fibrous tissue may be seen small trabeculae of primitive, poorly calcified new bone. The fibrous tissue appears relatively avascular. There may be seen small nests of giant cells, resembling osteoclasts. Occasional islands of hyaline cartilage may be found within the fibrous tissue. This is not a constant finding.

I wish to quote verbatim from Lichtenstein's paper in order to express his views on the pathogenesis of this disease: "The characteristic pathologic feature of polyostotic fibrous dysplasia appears to be a disturbed function or development of the bone-forming mesenchyme, which results in replacement of the spongiosa and filling of the medullary cavity of affected bones by fibrous tissue in which trabeculae of poorly calcified primitive new bone are developed by osseous metaplasia. The seemingly complex histologic picture becomes much easier to interpret if one predicates the multipotential capacity of this undifferentiated fibrous tissue. The latter normally gives rise to the spongiosa and to the myeloid or fatty marrow, but under pathologic conditions it may develop in several anomalous ways. By osseous metaplasia, it gives rise to osteoid and primitive fiber bone. By cartilaginous metaplasia, it gives rise to sporadic, isolated islands of hyaline cartilage, which tend to become calcified. By fibroblastic differentiation, it gives rise to mature collagenous connective tissue. Finally, by coalescence of its nuclei, it may give rise to multinuclear cells, indistinguishable from osteoclasts. Whatever stimulates the continued perverted activity of the undifferentiated fibrous bone-forming mesenchyme, or initiates the disorder remains a matter of conjecture. The clinical history of symptoms dating back to early childhood strongly suggests a congenital basis for this curious anomaly."

In Lichtenstein's series, serum calcium determinations ranged between 9.8 and 11 mg. per 100 cc. The serum phosphorus estimations showed no significant change. In three of his cases, the phosphatase was considerably increased; namely, 17, 18 and 22 Bodansky units. Lichtenstein feels that the increased phosphatase values in polyostotic fibrous dysplasia afford additional evidence to support the contention of Bodansky and Jaffe that the activity of the enzyme phosphatase is proportional to the stimulus to new bone

formation. It is interesting to note that the serum values of the patient reported in this paper were such as to confuse the diagnostic problem considerably. In Richmond, the serum calcium was 13 mg. and the phosphorus 2.8 mg. When these were repeated in Bernheim's laboratory at the New York Hospital, they were 11.3 mg. of calcium and 3.2 mg. of phosphorus. The phosphatase determination was four units.

In none of the cases studied at the Hospital for Joint Diseases were calcium balance studies made. In our own case, such studies showed an excretion of 1547 mg. of calcium in the urine and feces over a three-day period, during which a measured diet containing 300 mg. was ingested. This indicated a

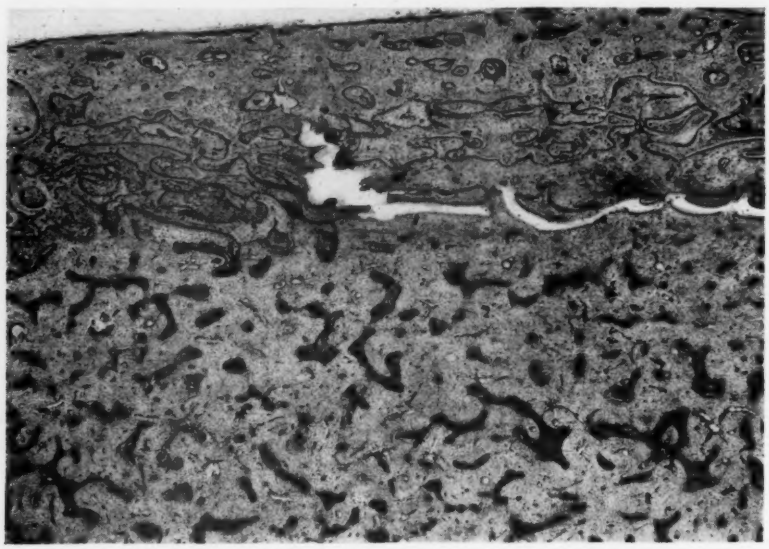

FIG. 12.—Microscopic section from a typical case of "polyostotic fibrous dysplasia," showing the fibrous tissue filling the medullary cavity and thinning of the bone cortex due largely to erosion of the endosteal surface. Dispersed irregularly in the fibrous tissue are small trabeculae of primitive poorly calcified new bone. (From the collection of Dr. Henry Jaffe, Hospital for Joint Disease, New York.)

marked negative calcium balance which, in conjunction with all other findings, was considered conclusive evidence of an existing hyperparathyroidism.

The importance of the laboratory and roentgenologic findings in Case 3 now becomes obvious. It is evident that a patient with polyostotic fibrous dysplasia may present all the clinical and confirmatory laboratory evidence usually associated with hyperparathyroidism, or osteitis fibrosa cystica.

## CONCLUSIONS

It is suggested, therefore, that the surgeon, when confronted with suspicious bone lesions evident in the roentgenograms, and serum estimations of calcium and phosphorus which are outside the normal limits, and in spite of the fact that calcium metabolism studies may show a negative balance, should not be too hasty to advise exploration of the neck for a parathyroid adenoma. It is suggested further that, when doubt exists as to the diagnosis, additional

investigation should be undertaken to clarify the situation. This consists, first, of roentgenologic examination of the skeleton to determine whether the bone lesions have a predominantly unilateral distribution and second, the performance of a bone biopsy. The latter will definitely establish the diagnosis by differentiating the characteristic histologic pictures of polyostotic fibrous dysplasia and hyperparathyroidism (Figs. 12 and 13).

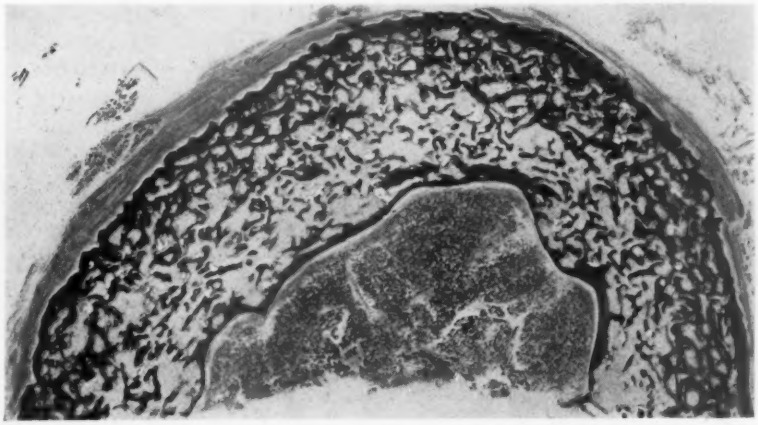

FIG. 13.—Photomicrograph from a typical case of osteitis fibrosa cystica which should be compared with Fig. 12. This indicates that most of the bone change has taken place in the cortex with cystic degeneration and bone absorption as the predominant features. There is little or no change in the medullary cavity. (From the collection of Dr. Henry Jaffe, Hospital for Joint Disease, New York.)

DISCUSSION.—DR. EMIL GOETSCH (Brooklyn) said that Doctor Garlock had drawn attention to some very interesting as well as anomalous conditions which simulate parathyroid dysfunction but which are not due to parathyroid hyperactivity. The present status of knowledge concerning the parathyroid is rather confused but, as with previous clinical syndromes, the time will come when those due to parathyroid dysfunction will be more clearly differentiated.

The first two cases were unusually illustrative of the clinical conditions typical of parathyroid adenomatous tumors. There were the usual blood changes with elevation of the calcium level and a diminution in the phosphorus content. There was an increase in the phosphatase and the bone changes first described by von Recklinghausen were typical of those found in instances of hyperactivity of the parathyroid. It is very satisfactory to have found parathyroid adenomata with the establishment of the real cause of the disease, and it is very satisfactory to have obtained such good results after the removal of the parathyroid tumors. Incidentally, too, the calcium and phosphorus content of the blood returned to normal, and there was an improvement in the condition of the bones due to redeposition of calcium in those areas from which calcium had been absorbed.

The third case was rather troublesome in that the bone changes were certainly suggestive of those found in hyperparathyroidism and there was associated a negative calcium balance, an association of findings very suggestive of parathyroid tumor. After most careful search, no parathyroid tumor or adenoma was found. Doctor Garlock called attention to the disease designated as "polyostotic fibrous dysplasia" which exhibits the findings just described and warned against being too hasty in operating for a supposed instance of

parathyroid tumor even in the presence of a negative calcium balance and rarefaction of the bones.

The last case further demonstrated the great value of calcium balance studies and that one cannot rely upon blood calcium findings alone. Calcium balance determinations over the three-day period are far more valuable than a few isolated blood calcium determinations. Thus, a high calcium change may be present due to failure of excretion or, on the other hand, a relativeiy low calcium may be present in instances of rapid excretion. The value of calcium balance determination, therefore, is obvious. Doctor Garlock warned against too prompt exploration of the neck before a very careful clinical investigation has been made. Too many conditions in which calcium deposits have been found, such as arthritic changes or arteriosclerosis, have been attributed to parathyroid dysfunction. Even scleroderma or arteriosclerosis has been attributed to disturbances of parathyroid function. In a recent case of this kind with extensive sclerosis, stiffness of the face and "wooden" fingers, Doctor Goetsch was prevailed upon to explore for parathyroid tumor but after most careful search nothing was found, although there was present an interesting generalized fibrous sclerosis, deep as well as superficial. Incidentally a small isolated tumor of thymus tissue was found at the right lower pole. At times it is difficult to identify, absolutely, parathyroid tissue for it may be atypical and thus be confused with the appearances commonly noted in true lymph nodes or hemolymph nodes commonly found in the region of the parathyroids. Also there may be an admixture of lymphoid tissue. Illustrations are sometimes exhibited which appear more like lymphoid tissue than true parathyroid tissue.

Dr. Henry L. Jaffe (New York) thanked Doctor Garlock for his generous acknowledgment of the work of Doctor Lichtenstein and himself in connection with "polyostotic fibrous dysplasia," the pathology of which they had been able to clarify on the basis of cases coming under their observation, which helped to establish it as an entity.

A number of points bear reemphasis. One rarely observes a clear-cut increase in the serum calcium value in "polyostotic fibrous dysplasia." Of the cases observed by Doctor Jaffe only two had figures above 11, and those figures were below 11.5 mg. per 100 cc. The serum phosphorus figures (which are also important in the diagnosis of true cases of hyperparathyroidism) were more or less normal in all the cases observed. The serum phosphatase activity values were high except in one case, and that was a man of 47. His biopsy specimen showed relatively little new bone in the fibrous tissue filling the marrow cavity and, furthermore, there was not very extensive involvement in his case. In younger subjects, with more extensive lesions, the phosphatase values were high and, indeed, as high as in hyperparathyroidism.

The marked negative calcium balance reported by Doctor Garlock threw Doctor Jaffe off altogether. On empiric grounds he could not believe that cases of "polyostotic fibrous dysplasia" should show pronounced negative calcium balances. In fact, he considered making calcium balance studies on several of his cases, but since the osseous involvement was not generalized and was often quite limited, he felt certain that the calcium balance studies would show nothing remarkable. It is true that Doctor Goetsch emphasized the value of calcium balance studies and many people have been stressing their value in diagnosis of various skeletal diseases. Nevertheless, Doctor Jaffe doubted the value of the information obtainable from a three-day calcium balance study as done by present methods. The fact is that a normal person on a diet containing 100 mg. of calcium shows in a three-days' balance study a negative calcium balance and the calcium balance studies only measure the degree of negativeness from person to person. Doctor Jaffe did not doubt Doctor Bernheim's figures for Doctor Garlock's case, yet he was skeptical

that subjects with "polyostotic fibrous dysplasia" show pronounced negative balances. He emphasized that it is of the utmost importance to clarify this point in the differential diagnosis between this condition and hyperparathyroidism. Certainly if studies of additional cases show that the calcium balance is severely negative, then there is another confusing point in the differential diagnosis between it and hyperparathyroidism. Doctor Jaffe hoped for the sake of diagnosis that it would prove to be otherwise.

Regarding roentgenologic findings, Doctor Jaffe affirmed Doctor Garlock's statement that there is a tendency for the lesions to be unilateral. Furthermore, the lesions, statistically, tend to concentrate in the femur and tibia, although he had seen them in the calvarium and particularly in the occipital bone, also in the ribs, in the vertebrae, in the pelvic bones, and even in the small bones of hands and feet. The reason why cases of "polyostotic fibrous dysplasia" are misdiagnosed as hyperparathyroidism is that the fibrous tissue growing in the marrow cavity erodes the inner surface of the cortex and may even distend the bone, and, of course, the bone shadow is suggestive of a cyst. Actually these bones are not cystic. When one penetrates the cortex, one finds the marrow cavity filled by a hard and almost rubbery tissue. Even where the picture is, roentgenographically, clearly that of a cyst, this type of tissue exists.

One important point in the roentgenologic differential diagnosis between "polyostotic fibrous dysplasia" and hyperparathyroidism is rarefaction of the cortex of most bones in the latter and the absence of cortical rarefaction except in the affected bone in the former.

Dr. Carl G. Burdick (New York) asked Doctor Jaffe regarding the outcome of some of the cases he had followed.

Doctor Jaffe answered that the first case he saw was a girl who at the time of his first observation (1926) showed involvement of a number of ribs, one femur, and one tibia. She had a fracture through the neck of the femur. She left the Hospital for Joint Diseases and some years later Doctor Jaffe traced her to Long Island College Hospital where a bone graft had been inserted in the femur. She became a mother of two children and her illness was not particularly aggravated by the pregnancies. She still has the disease and presumably she will go on having it. Some of the lesions may subside somewhat as she becomes older. The fibrous tissue may become more collagenous and the condition consequently less progressive.

Dr. Bradley L. Coley (New York) recalled a case of malignant tumor of the parathyroid gland which he showed before the New York Surgical Society in 1936. At that time it was assumed that the changes in the patient's spine, which originally brought him under Doctor Coley's care, were due to hyperparathyroidism associated with the tumor. The subsequent course, however, proved that the lesion in the spine was really a metastasis, and when last seen the patient was growing rapidly worse. In retrospect, the case represents one of malignant tumor of the parathyroid (adenocarcinoma of parathyroid origin) with metastasis to the lumbar spine.

Dr. John H. Garlock (New York), in conclusion, said that Doctor Jaffe's remarks about calcium balance studies were very well taken. Nevertheless, he could not alter his belief in Doctor Bernheim's figures. Doctor Bernheim has done many calcium balance studies over a long period of time in her own laboratory in New York Hospital, and also has a technician who has been working under her for many years, and knows how to carry out the studies. In the case presented these studies definitely showed a marked negative calcium which Doctor Garlock considered to be a very important finding.

# THE TREATMENT OF POSTOPERATIVE TETANY WITH DIHYDROTACHYSTEROL*

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THE parathyroids were first recognized as independent organs by Sandstroem in 1880,[1] but Gley[2] (1892) was the first to show that their removal was associated with the production of tetany. Two outstanding facts have long been known regarding the physiology of the parathyroids as glands of internal secretion: (1) Their influence on calcium metabolism; and (2) the specific relation of hypoparathyroidism to altered irritability of the nervous system, these two relationships being closely associated.

Excision of the parathyroid glands of dogs produces, after a latent period of variable length, an acute tetany, which is characterized by a hypocalcemia. The symptoms may be directly controlled to a greater or less degree by means of (a) injections of extracts of the parathyroid glands (Beebe,[3, 4] and Berkeley and Beebe[5]); (b) by dietary control in conjunction with the administration of calcium[6, 7]; (c) symptomatically by means of narcotics and hypnotics.[8] The effectiveness of such therapy suggested that the function of these glands was the regulation of calcium metabolism, *i.e.*, that they control the absorption and retention of calcium as shown by MacCallum and Voegtlen.[9] However, other metabolic changes, which might favor the development of convulsions following parathyroidectomy, likewise occur. These changes, including depressed liver function (Carlson and Jacobson[10, 11]), the appearance of guanidine, histamine and similar toxic bases in the urine (Koch[12, 13]), diminished carbohydrate tolerance (Underhill and Saiki[14]), are probably of secondary concern.

The basic importance of the physiology of the parathyroid glands is due to their interrelationships with other glandular structures, the fundamental relation of calcium metabolism to cellular permeability and function, and from a clinical standpoint to their relation to idiopathic and postoperative tetany. The latter condition develops to a greater or lesser degree following either (a) the accidental removal of one or more of these structures during the course of a thyroid operation, or (b) their destruction through interference with their blood supply, postoperative infection, or scar tissue compression. Under such conditions, postoperative tetany is characterized by a hypocalcemia of greater or lesser degree. The severity of the symptoms parallels as a rule the degree of hypocalcemia, but rather wide individual variations are to be noted in different patients. However, restoration of normal values for a particular patient results in a complete disappearance of symptoms of tetany,

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during the period in which a normal calcium balance is maintained or certain minimum blood calcium values are exceeded.

The normal blood calcium and phosphorus values vary from 9 to 11 mg. and from 3.2 to 4.5 mg. per 100 cc. of blood serum, respectively. As a rule, symptoms of tetany become manifest when the serum calcium values fall to, or below, 7 mg., while the serum phosphorus values on the other hand usually rise. It is quite impossible, however, to establish an arbitrary tetany level for calcium values. Symptoms in some patients may not develop until a value of 6.8 mg. has been reached, while in others, similar symptoms may be pronounced with serum calcium values of 8.1 to 8.3 mg.

The therapeutic correction of clinical hypoparathyroid tetany may be largely accomplished in one or more of several ways, *i.e.*, the administration of milk and calcium by mouth in conjunction with calcium salts by vein; the administration of parathormone by injection, together with calcium salts; the oral administration of vitamin D; or surgically by the implantation of parathyroid gland tissue.

The treatment of tetany with calcium alone, irrespective of the route of administration, is usually unsatisfactory due to the transient character of such therapy and incomplete relief obtained. This is due to the fact that the calcium salts, when administered orally, are irregularly absorbed from the gastrointestinal tract, and owing to poor retention (rapid urinary and fecal excretion), medication must be repeated at short intervals.

The administration of parathormone or vitamin D presupposes that a calcium-rich diet or calcium salts *per se* are used in conjunction with these agents so that an improved absorption and retention of calcium is possible. Neither of these agents alone, however, is entirely suitable for the treatment of an acute attack of tetany, in that reactions are demonstrable only after a latent period and the peak-effects occur only after nine to 12 and 36 to 48 hours, respectively. Consequently, control of clinical tetany with these agents requires careful consideration of the latent period of each agent in relation to symptomatology, as well as the total period of its effects. Another factor of clinical and, particularly, economic importance is that the patient gradually develops a tolerance to the effects of parathormone so that eventually a unit blood calcium change requires progressively larger dosages.[15, 16]

The surgical treatment of hypoparathyroidism is of temporary value only in that the degree of relief is unpredictable and the efficacy of such therapy under optimal conditions progressively diminishes after a variable period of time due to atrophic changes in the implanted tissue.

In any event, although the diagnosis of frank postoperative tetany is readily made, the establishment of the degree of an acute attack or the presence of latent tetany can be detected only by determinations of the serum calcium values. We consider the routine determination of blood calcium values both pre- and postoperatively in each patient subjected to thyroid surgery as of primary importance, for both diagnostic purposes and therapeutic control.

In the treatment of tetany, immediate alleviation of symptoms is the first

consideration at all times, but *prolonged* effective control of the tetany that maintains normal level of serum calcium, with as little discomfort to the patient as possible, is the goal sought for. We wish to report that we have used a new therapeutic agent which will definitely control tetany. This substance is a sterol—dihydrotachysterol—perfected abroad by Holtz and his associates and known as Anti Tetany 10 ("A. T. 10").*

Holtz[17] and his coworkers, von Brand,[18] Schreiber,[19] and Isemer and Stichnoth,[20] under the direction of Professor Windaus and his collaborators, Hess,[21] Lüttringhaus,[22] and Holtz,[23] were among the first to observe that very large doses of "Vigantol,"[24, 25] a German preparation of irradiated ergosterol, led to a series of toxic symptoms, designated by some authors as hypervitaminosis D. Hypervitaminosis D has as one of its characteristic features a hypercalcemia. The main symptoms of this condition include headache, nausea, vomiting, hematuria and a moderate elevation of blood pressure. An abnormally high calcium level in the serum, over a relatively long period of time, results in the deposition of calcium salts in the heart, blood vessels, kidneys and other organs of the body.

Holtz searched for a toxic factor as a possible cause of the hypervitaminosis and found that by prolonging the irradiation of ergosterol under certain conditions he could destroy, in large part, the antirachitic factor, vitamin D, but that the "calcinose" factor was still present. Extensive investigations of the action of the "calcinose" factor by Holtz and his associates[26, 27, 28] were undertaken on both normal and parathyroidectomized animals. The results indicated that this "calcinose" factor caused a marked increase in the amount of calcium in the serum of normal animals with a deposition of calcium in the organs of the body; in the parathyroidectomized animals appropriate amounts of this "calcinose" factor elevated the serum calcium to normal and prevented the symptoms of parathyroid insufficiency. Similar results were obtained in our series of patients following the development of postoperative hypoparathyroidism.

Holtz,[29, 30, 31] and then Holtz, Gürsching and Kraut,[32] described this preparation which they called "A. T. 10" and which is now designated chemically as dihydrotachysterol. This compound, although derived from irradiated ergosterol, differs from the parent substance in that it is almost devoid of any antirachitic† action, while the calcium mobilizing factor persists and exceeds that of viosterol.

Crude irradiated ergosterol, according to our present knowledge (Fieser[33] and Friedmann[34]), contains at least four sterols, *i.e.*, tachysterol, lumisterol, dihydroergosterol, or vitamin $D_2$ (calciferol), suprasterol, and pyrocalciferol.

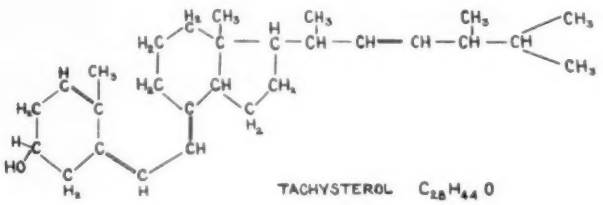
* This material has been supplied to us through the courtesy of the Winthrop Chemical Company for experimental purposes.

† Personal communication from Dr. O. W. Barlow: "The antirachitic action of 'A. T. 10' is approximately 1/600 of that of an equal amount of crystalline Vitamin $D_2$." We take this opportunity to thank Doctor Barlow for his many helpful suggestions and criticisms in the preparation of this manuscript.

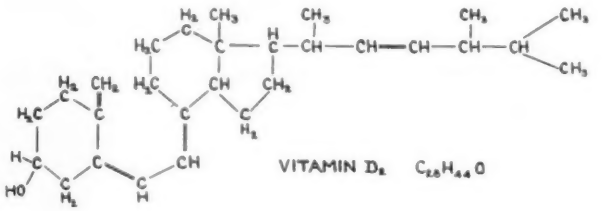
Attention is directed to the fact that calciferol contains only one sterol. Müller[35] states that tachysterol and vitamin $D_2$ (calciferol) yield identical dihydro derivatives, and consequently possess the same carbon skeleton. However, the rather striking difference in the physiologic effect of these two compounds strongly indicates that some difference exists between the points of hydrogenation of the dihydrovitamin ($D_2$) and dihydrotachysterol.

The following formulae show the structural relationship between the various sterol fractions mentioned in the development of dihydrotachysterol.

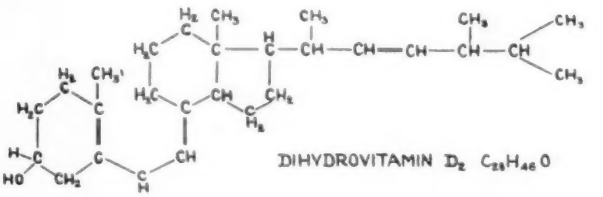
FORMULAE SHOWING THE STRUCTURAL RELATIONSHIPS BETWEEN THE VARIOUS STEROL FRACTIONS IN THE DEVELOPMENT OF DIHYDROTACHYSTEROL ("A.T. 10")



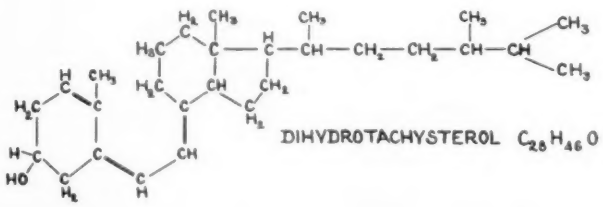
TACHYSTEROL $C_{28}H_{44}O$



VITAMIN $D_2$ $C_{28}H_{44}O$



DIHYDROVITAMIN $D_2$ $C_{28}H_{46}O$



DIHYDROTACHYSTEROL $C_{28}H_{46}O$

The excellent clinical results that have been reported in the literature,[16, 36, 37, 38, 39, 40] following the use of "A. T. 10" in various conditions in which disturbances of calcium metabolism were present, stimulated us to make clinical tests with this new calcinose factor. Our experiences to date have been of such nature as to merit reporting. These include the oral

administration of the product in the form of a 0.5 per cent sesame oil solution of dihydrotachysterol to a series of five patients over the past two and one-half years. The subjects include three chronic and two acute postoperative tetanies following surgery of the thyroid glands performed by various surgeons. The courses of two of these five subjects, one acute and one chronic case of tetany, are herewith reported.

**Case 1** (Chart 1).—Female, age 24, with a diagnosis of hyperthyroidism. Basal metabolism +31.

April 21, 1936: A subtotal thyroidectomy was performed. *Pathologic Examination.*—Dr. Fred D. Bullock: "Parenchymatous and colloid goiter. It is interesting to note that imbedded in one of the lobes there is a fairly normal parathyroid."

April 22: Thyroid crisis; temperature 108° F.; symptoms of tetany of a very severe form.

May 4: Calcium was 5.9 mg. Patient was given 50 units of parathyroid hormone and calcium gluconate, one ounce three times daily, with viosterol ten drops t.i.d. From the third to fifth week, she received 50 units of parathyroid hormone daily, and the calcium varied from 5.9 to 8.7 mg.

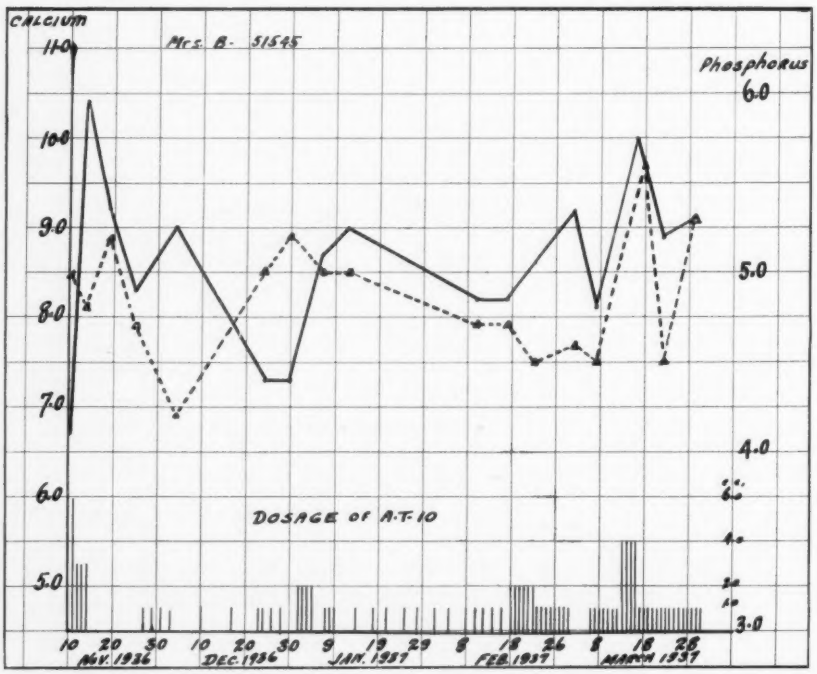
May 11: Calcium was 5.9 mg.

May 18: Calcium was 6.9 mg.

May 21: Basal metabolism —10.

Patient received parathyroid hormone until she was discharged, May 23, 1936.

Chart 1.




It will be noted that in spite of treatment with parathyroid hormone and calcium salts, at no time was the calcium maintained anywhere near normal

level. After the patient had been discharged from the hospital, she would have to come in occasionally at night, with severe symptoms of tetany, in order to receive injections of parathyroid hormone. A positive Chvostek sign was present continuously. Patient had complained of tingling, sleeping of the hands and legs, and was more or less uncomfortable.

In the fall of 1936 we received a supply of "A. T. 10," and the patient returned to the hospital to be treated for her tetany (Chart 1).

November 10, 1936: On admission her calcium was 6.7 mg., phosphorus 5.0 mg. Patient was given 6 cc. of "A. T. 10" by mouth with 45 gm. of calcium gluconate divided into three doses.

November 11: 3 cc. "A. T. 10" and calcium gluconate.

November 13: 3 cc. "A. T. 10."

November 14: 3 cc. "A. T. 10." Calcium rose to 10.4 mg. and the phosphorus was 4.8 mg. At the end of 48 hours, after the "A. T. 10" was given, all symptoms of tetany had been alleviated with the exception of a mild Chvostek sign. For the next three days, no medication was given and the calcium fell to 9.2 mg. with a rise in the phosphorus to 5.2 mg.

November 20–25: No "A. T. 10" was given, but 45 Gm. of calcium gluconate per day.

November 25: Calcium fell to 8.3 mg., phosphorus being 4.7 mg. Patient was discharged with marked improvement in her tetany and was to receive ambulatory treatment.

December 4: Patient was given 1 cc. of "A. T. 10" every other day plus 45 Gm. of calcium gluconate daily. Calcium rose to 9 mg. and the phosphorus was 4.2 mg.

January 24, 1937: Patient received 1 cc. "A. T. 10" once a week and no calcium gluconate.

December 19: Patient experienced a severe shock due to the fact that her baby had been scalded to death. It is remarkable to note that in spite of this she developed no symptoms of tetany.

December 23: Calcium was 7.3 mg., phosphorus 5 mg. The dosage of "A. T. 10" was increased 1 cc. every other day plus the calcium gluconate.

December 30: Calcium was 7.3 mg.

For four days, up to January 5, 1938, she received 2 cc. per day plus the calcium gluconate, and January 6, the calcium rose to 8.7 mg., phosphorus 5 mg.

January 7, 8, 9: She received 1 cc. daily plus the calcium gluconate.

January 10: Calcium was 9 mg., phosphorus 5 mg.

February 9: During preceding three weeks, the patient received 1 cc. "A. T. 10" twice a week plus calcium gluconate daily.

February 10: Calcium was 8.2 mg., phosphorus 4.7 mg. From this period on, the amount of calcium gluconate was reduced to 1 Gm. three times a day.

February 17: Calcium 8.2 mg., phosphorus 4.7 mg.—no change.

February 18–23: Patient received 2 cc. of "A. T. 10" daily plus 1 Gm. calcium gluconate t.i.d.

February 25: Calcium was 8.6 mg., phosphorus 4.5 mg.

March 1: Patient received 1 cc. "A. T. 10" daily.

The fall in calcium without any symptoms of tetany and a very greatly diminished Chvostek sign were rather disturbing elements. Examination disclosed that the patient was now two months pregnant. She had not been ill with other pregnancies, but at present was very ill each day with nausea and vomiting.

"A. T. 10" was now increased to 1 cc. daily for one week, and on

March 2: Calcium was 9.2 mg., phosphorus 4.6 mg.

March 3, 4, 5, 6: No "A. T. 10" but calcium gluconate.

March 7: Calcium 8.1 mg., phosphorus 4.5 mg.; patient received 1 cc. "A. T. 10" every day for a week. She was admitted to the hospital in March, 1937, and was given 1 cc. "A. T. 10" every day with calcium gluconate for a week.

March 13, 14, 15, 16: Patient received 4 cc. of "A. T. 10" plus calcium gluconate.

March 16: Calcium rose to 10 mg., phosphorus 5.6 mg.

March 16: On advice of gynecologists, a hysterectomy was performed. No signs or symptoms of tetany occurred postoperatively.

March 17, 18, 19: Patient had received, two to three days after operation, 10 cc. calcium gluconate intravenously.

March 19: Patient received 1 cc. per day of "A. T. 10" plus calcium gluconate.

March 22: Calcium 8.9 mg., phosphorus 4.5 mg.

March 29: Patient discharged. She had received 1 cc. of "A. T. 10" daily, plus calcium gluconate.

March 29: Calcium 9.1 mg., phosphorus 5.3 mg.

*Comment.*—It is interesting to note that "A. T. 10" alone apparently does not work as well as when given in conjunction with calcium salts; that the dosage at first was rather large—6 cc.—and was decreased as soon as symptoms of tetany were lessened. However, we have also observed that the calcium rises slowly and when once the symptoms of tetany are alleviated, small doses are sufficient to maintain a normal serum calcium level, and that with any decrease in the blood calcium the dosage is somewhat increased or given over a longer period of time. The object, of course, is to get the patient symptom free and to maintain a normal calcium level with as little "A. T. 10" as possible. This patient will probably have to take "A. T. 10" for a long period of time, with gradual reduction in the dosage until a normal calcium balance is established. But this is of no particular inconvenience to the patient as the substance is tasteless and, of course, given by mouth. The patient has been symptom free ever since she has taken the "A. T. 10."

**Case 2** (Chart 2).—Female, age 32, was first seen in May, 1936, and complained of nervousness, palpitation, and loss of weight. One miscarriage six years ago was followed by loss of about 40 pounds in weight which was never regained. General health good. She was admitted to the hospital May 25, 1936.

May 26: Basal metabolism +65.

June 2: Basal metabolism +73.

June 8: Basal metabolism +49.

June 22: Patient operated upon. Subtotal thyroidectomy. *Pathologic Examination.*—Dr. F. D. Bullock: Colloid and parenchymatous goiter.

June 25: Patient developed twitching of the legs and other signs of beginning tetany.

June 26: Blood calcium was 4.9 mg., phosphorus 5.7 mg.

June 27: She received 10 cc. "A. T. 10" and 45 Gm. calcium gluconate.

June 28: Same dosage.

June 29: Blood calcium was 5 mg., phosphorus 6.1 mg. Symptoms of tetany are considerably diminished.

June 30: Calcium 5.4 mg., phosphorus 6.2 mg.

July 1, 2: Patient given 4 cc. "A. T. 10" and 45 Gm. calcium gluconate.

July 3: Calcium 5.8 mg., phosphorus 6.4 mg. Very little change.

July 3, 4, 5: Patient received 1 cc. "A. T. 10" and 45 Gm. calcium gluconate.

July 7: Calcium 6.5 mg., phosphorus 4.9 mg.

July 6, 7: "A. T. 10" discontinued, and only calcium gluconate given.

July 8: One cubic centimeter "A. T. 10" and 45 Gm. calcium gluconate.

July 9: No "A. T. 10" and only calcium gluconate given.

July 10: One cubic centimeter "A. T. 10."

July 11: Calcium rose to 7.1 mg., phosphorus 3.9 mg.

July 13: One cubic centimeter "A. T. 10."

July 14: Calcium 7.1 mg., phosphorus 4.6 mg. No symptoms of tetany, with the exception of a slight cramp in the leg and a mild Chvostek sign. Patient discharged. Basal metabolism +14. Condition good. Ambulatory treatment to be continued.

July 14 to August 5: Patient given 1 cc. "A. T. 10" twice a week for one week only, plus 45 Gm. of calcium gluconate.

August 5: Calcium 8 mg., phosphorus 4.5 mg. Still a mild Chvostek sign.

August 4, 1936, to January 13, 1937: Patient had been placed on calcium gluconate plus calcium phosphate with a little viosterol, with the view to seeing whether serum calcium would remain above the tetany level *without* the use of "A. T. 10." However, serum calcium during this period, on September 3, was 9 mg. and on November 18, 8.2 mg.

January 13: Calcium had fallen to 8 mg. with phosphorus 4 mg., and it was decided to give the patient some "A. T. 10" to stimulate calcium mobilization. Patient received "A. T. 10," 1 cc. daily, for ten days with calcium gluconate.

January 26: Calcium was 8.3 mg., phosphorus 3.4 mg. Dosage was continued. Chvostek sign was very mild. Patient feels well; no complaints.

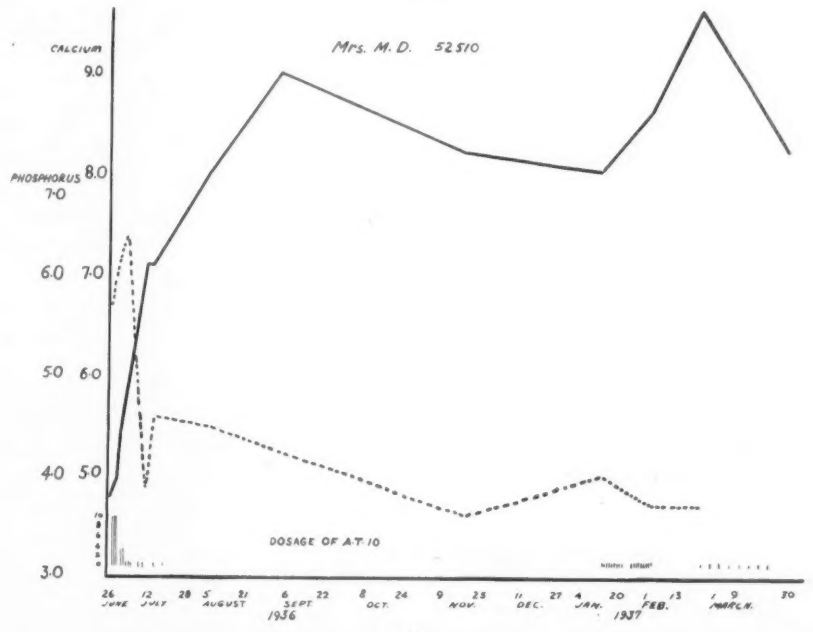
January 26: Calcium was 8.6 mg., phosphorus 3.7 mg.

February 23: One cubic centimeter "A. T. 10" was given twice a week.

February 25: Calcium was 9.6 mg., phosphorus 3.7 mg.

March 30: Calcium was 8.2 mg. No complaints, excellent condition. Chvostek sign has practically disappeared.

CHART 2.



*Comment.*—This case of acute tetany, in which we were fortunate to start immediate treatment with "A. T. 10," is of particular interest because it brings out the fact that while the blood calcium was slow in rising, her symptoms of tetany were alleviated in a very short time after administration of the "A. T. 10." Within 18 days, there was a marked rise in the blood calcium which was sustained with a comparatively small dose of "A. T. 10." During the period in which medication with the sterol was discontinued, there was a sustained normal calcium level. Of course, the question here arises as to the cumulative action of this preparation. Whether this action actually exists or not is, in our minds, open to question because as soon as "A. T. 10" is discontinued, there may be a fall in the blood calcium. But it can be very easily maintained by small dosages over a period of weeks. We are studying this question further. This patient has been symptom free, that is of actual tetany, ever since she has been taking "A. T. 10," and on the last examination was in excellent condition. This case demonstrates again the absolute necessity of determining the blood calcium and phosphorus after each period of treatment. It is only in this manner that marked deviations from normal can be observed and the figures will act as a guide for the amount of "A. T. 10" to be given.

## CONCLUSIONS

This new therapeutic agent, dihydrotachysterol ("A. T. 10"), has great value in the control of postoperative tetany, and the symptoms can be alleviated within 48 to 72 hours after instituting treatment. These patients are usually irritable, extremely nervous and physically played out, and certainly the removal of these symptoms, as soon as possible, is of the utmost necessity.

Therapy with dihydrotachysterol must be strictly individualized, *i.e.*, the blood calcium level must be carefully checked at intervals before and after medication has been instituted. Until the maintenance dosage has been determined, the amount of "A. T. 10," given by mouth, may vary from 1 to 3 cc. every other day to 1 to 2 cc. per week, depending on the severity of the deficiency. We have on occasion administered dosages as high as 10 cc. daily for a period of several days, but during this period have kept a careful check on the patient's serum calcium findings. The product is not harmless and excessive dosage may well induce symptoms of hypercalcemia.

A plea is made for the routine determination of serum calcium both pre- and postoperatively in all thyroid surgery.

This new substance in our hands has been of definite value both from a subjective and objective standpoint in the control of tetany. These observations, taken in conjunction with those of Holtz and his coworkers, would suggest that dihydrotachysterol merits further chemical study and it is recommended for the treatment of postoperative hypoparathyroid tetany.

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Discussion.—Dr. DeWitt Stetten (New York) felt that there was little left for him to say except to confirm in every detail what Doctors Pickhardt and Bernhard had reported so well. He was fortunate enough to have had the opportunity of observing, at first hand, some of their work, as two of the patients on whom they have reported were from his service. One of them had been shown and there was no need for him to make any further comment about her.

One of the patients to whom they had referred was a woman, age 60, upon whom Doctor Stetten had performed a left lobectomy for a large colloid goiter, October 24, 1929, and upon whom he again operated, October 17, 1933, for a similar condition in the right lobe, when a resection of part of the right lobe was undertaken. About three weeks after this second operation she developed definite, rather severe symptoms of tetany, the serum calcium dropping as low as 4 mg. per 100 cc. Only with very large doses of calcium in the form of calcium gluconate, mainly intravenous, and intramuscular injections of parathormone, was she kept moderately comfortable—relieved but never well. Her calcium could never be raised much above 6.5 mg., and the phosphorus rose as high as 6 mg. At times it was necessary to give her, almost as an emergency, an intravenous injection of calcium gluconate, and

although the tetanic spasms were relaxed promptly, they recurred often within a few hours. She managed to worry along for three years until Doctors Pickhardt and Bernhard began this work at Lenox Hill Hospital, when, on December 2, 1936, Doctor Stetten persuaded her to enter the hospital for a trial of this new agent. She did so, much against her will as she objected to further experimentation, but after a few days of treatment she was beaming and has never been able to thank him enough for having talked her into giving this drug a trial. She is really cured of her tetany, as shown not only by the complete and persistent disappearance of the carpopedal spasms, Chvostek and Trousseau signs, but also by a serum calcium that is practically normal, and the characteristic drop in the blood phosphorus—and that with only relatively small doses of the drug by mouth at comparatively long intervals, supplemented by moderate amounts of calcium also administered orally. She has even been able to resume her occupation as dressmaker.

Whatever "A. T. 10" or dihydrotachysterol is or in whichever way it produces its result, it is certainly a calcium fixative, and, as such, apparently controls postoperative parathyroprival tetany. The effect of the administration of this agent in this distressing disturbance is most dramatic—really little short of miraculous—and Doctor Stetten does not think he is exaggerating when he states that, in its more limited field of application, it is only comparable to the specific actions of insulin in diabetes or liver extract in pernicious anemia.

Doctor Stetten feels that the Society is under a great debt to Doctors Pickhardt and Bernhard for introducing this drug into America, for so carefully studying its action, and for so convincingly presenting to it this evening, the results of their research.

# CONGENITAL DIAPHRAGMATIC HERNIA*

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DIAPHRAGMATIC hernia is a protrusion of the abdominal organs into the chest through a defect in the diaphragm. In the congenital type, with which this paper deals, the defect is usually caused by a failure of fusion in the various parts of the embryonic diaphragm, and the herniated organs may lie free in the chest with or without a peritoneal sac. Infrequently, herniae occur after fusion has taken place when the increase in the abdominal pressure causes a protrusion through some weak area. In the latter type, a peritoneal sac is more likely to be present than in the former.

It is still difficult to estimate the incidence of congenital diaphragmatic hernia because of the variance of published reports. In 1610, Ambroise Paré reported two traumatic cases. In 1769, Morgagni wrote the first monograph on the subject. In 1920, MacMillan[18] found only three cases among 15,000 roentgenologic examinations, and in 1924, Pancoast and Boles[22] reported 16 cases among 9,000 gastro-intestinal examinations. In 1931, Hedblom[12] collected 1,435 cases from the literature. In 1933, Dickson[7] reported 206 cases during the preceding 14 years from the roentgenologic department of the Toronto General Hospital. Harrington,[11] in 1933, stated that they had had five times as many cases at the Mayo Clinic during the preceding eight years as they had had during the previous 24 years. The purpose of this paper is to record ten cases of the congenital type from the Babies Hospital, New York, seen during the past seven years.

The most common sites of the defects in the diaphragm through which the abdominal organs pass are: The foramen of Bochdalek; the foramen of Morgagni; the dome of the diaphragm; and the esophageal hiatus. It is interesting to note that a hernia through the aortic or vena caval openings in the diaphragm has never been reported.

*Embryology.*—The diaphragm is so complex in development that it is not difficult to understand how a defect may arise where the different segments meet. The embryonic diaphragm consists of two parts: a ventral part which is the cephalic portion of the septum transversum, developing in the cervical region, and a dorsal part which is the pleuroperitoneal membrane, developing from the lateral body walls and destined to become the closing membrane between the pleural and the peritoneal cavities. The muscular portion of the diaphragm develops while it is in the cervical region from the third and fourth cervical myotomes on each side. During development, the diaphragm migrates from the region of the third cervical vertebra to its final location opposite the

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twelfth thoracic vertebra, and, during this migration, its plane of direction changes many times. It is thought that the original communication between the pleural and peritoneal cavities closes about the third month of intrauterine life. If this communication remains open, there is formed the pleuroperitoneal hiatus known as the foramen of Bochdalek. Since the liver lies over the right foramen, herniae at that point occur less often than on the left side. Failure of fusion of the costal and sternal fibers at either side of the sternum results in the formation of the foramina of Morgagni. It is more difficult to explain the defects in the dome of the diaphragm from an embryologic standpoint, other than to say that they are due to a failure of fusion of the elements at that point. The defects in the dome are more common on the left side. Hernia through the esophageal hiatus has been attributed either to the failure of development of the diaphragm at that point, or to the failure of migration of the stomach because of a short esophagus. These herniae usually have a peritoneal sac.

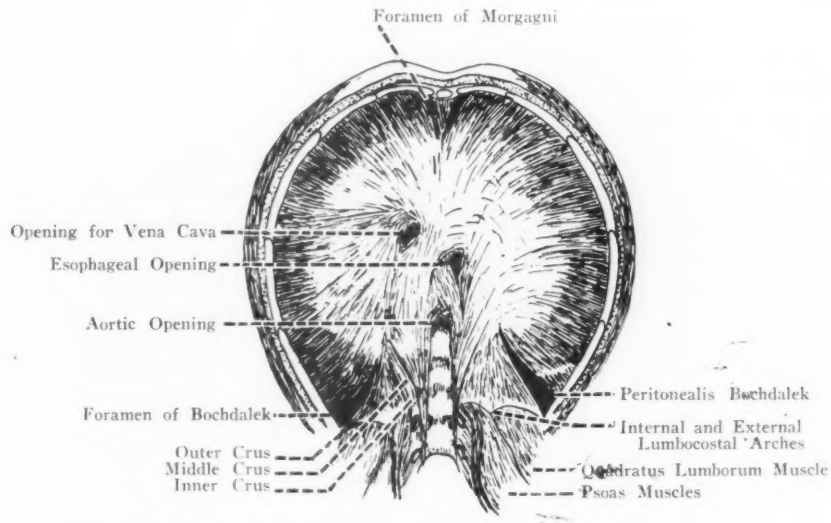


Diagram showing the location of the more common defects in the diaphragm. (Reproduced from Hermann, W. G.: Radiology, 22, 241–246, February, 1934.)

*Symptomatology.*—The symptoms of diaphragmatic herniae, which are the result of mechanical interference with the functions of the herniated viscera and of interference with the respiratory and circulatory organs upon which they encroach, may be respiratory, gastro-intestinal or a combination of both. If the herniated viscera press upon the heart or mediastinum, they may produce cyanosis, dyspnea and palpitation. If they consist of large or small intestine, nausea, vomiting or an intestinal obstruction may result from the crowding or kinking of the intestine. The symptoms vary to such a marked degree that the possibility of diaphragmatic hernia should always be kept in mind in cases exhibiting perplexing upper abdominal, respiratory or cardiac symptoms,

the etiology of which is difficult to ascertain. Many cases are symptom-free, and the hernia is discovered during the course of routine roentgenologic examination. One patient, whose condition was discovered after taking a roentgenogram of her chest, had been so well that it was difficult to convince her parents that a serious condition existed. Shortness of breath during the hot days of July was the only complaint of another. One patient suffered from loss of weight and vomiting since birth, while another had had a cough, diarrhea and tarry stools for several weeks.

*Diagnosis.*—The diagnosis of diaphragmatic hernia is greatly facilitated by roentgenologic examination. A roentgenogram of the chest will often show gas bubbles in the place of normal lung and frequently a displacement of the heart. Previous to 1900, when opaque media were first used to roentgenograph the gastro-intestinal tract, most of the diaphragmatic herniae were discovered at autopsy. Roentgenologic examination probably has been the greatest factor in the improvement in diagnosis and treatment of these cases. Not only does it identify the herniated structures, but it often demonstrates the location of the defect in the diaphragm.

*Time for Operative Intervention.*—Operative repair is practically the only relief for this condition. If the herniated contents contain any portion of the small or large intestine, operation should be advised immediately, especially in young children who evidence symptoms of impending obstruction, which is the most urgent indication. We were unfortunate enough to have a five weeks old baby (Case 4) develop a high intestinal obstruction while we were attempting to improve its general condition preparatory to operation. Another reason for not deferring operation is that, if the abdominal contents are allowed to remain in the chest for a long period, the development of the abdomen will be so retarded that it may not be large enough to accommodate the structures brought down into it from the chest. We had this experience with one of our patients (Case 5).

*Operative Approach.*—Opinions differ relative to the surgical approach in these cases. However, I prefer the abdominal approach, and have employed a subcostal incision in each case. In one instance of recurrence, it was necessary to make a thoracic incision in addition to the subcostal. The subcostal incision gives an excellent approach to the diaphragm, and it has always seemed easier to me to reduce the structures by traction from below than by forcing them down from above. The abdominal approach also lessens the occurrence of thoracic complications, and it gives one an opportunity to rearrange the abdominal contents to as near their normal position as possible. There are many advocates of the thoracic approach, notably Truesdale,[26] who recommends it in all cases. Harrington prefers the abdominal approach, and Hedblom believed that both the thoracic and the abdominal approaches had their special advantages and limitations, depending upon the anatomic and clinical type of hernia.

My experience with this small group of cases leads me to believe that it is desirable to expose the phrenic nerve in the neck before making the abdominal incision so that the nerve may be crushed if paralysis of the diaphragm seems advantageous after the hernia has been exposed. If the thoracic approach is employed this procedure is unnecessary, because the nerve may be crushed in the thorax. Positive pressure anesthesia is decidedly advantageous, and it was employed in most of my cases.

I believe these patients should be kept in an oxygen tent for several days after operation. Not only does the oxygen decrease the respiratory effort, thereby saving the child's energy, but the tent decreases the chances of respiratory infection because the temperature can be so easily regulated. Pleural effusion may follow operation, and if it occurs, the chest should be aspirated as often as the quantity of fluid demands.

The difficulties of the operation depend upon the location of the defect and the presence of adhesions, while the mortality depends to a large extent upon the age of the patient. Hedblom found that in 210 cases under one year of age, 75 per cent died before they were one month old. All but one of the operated cases here reported were under six months of age.

## Case Reports

**Case 1.**—P. N., male, age 4½ months, was admitted to the Babies Hospital, October 11, 1930, with the history of having had difficulty in breathing during the hot weather. He had had a convulsion 24 hours before admission. Otherwise, he was perfectly well. Roentgenologic examination showed small intestine in his left chest.

*Operation.*—The abdomen was opened through a left subcostal incision which revealed a hernia through the left foramen of Bochdalek. His chest contained all of the small intestine beyond the duodenojejunal juncture, all of the colon to a point beyond the splenic flexure, and the spleen. The splenic flexure of the colon was adherent to the parietal pleura, and it had to be mobilized before it could be brought down. All structures were, however, reduced, and the diaphragmatic opening was closed with mattress sutures of black silk, the edges of the hiatus being overlapped. The patient was kept in an oxygen tent and had a continuous intravenous drip of saline for several days postoperative. His convalescence was uneventful.

*Follow-Up.*—Six and one-half years postoperative, the child was normal in every way.

**Case 2.**—S. D., female, age 6 months, was admitted to the Babies Hospital, November 13, 1931, with a history of vomiting during the preceding five days. Full term birth by cesarean section. She had gained steadily, and had had no illness previous to the present one. She was a well developed, well nourished, white female child. A gastro-intestinal series showed small and large intestine in the right chest.

*Operation.*—November 20, 1931: The abdomen was opened through a right subcostal incision. On retracting the liver from the diaphragm, a patent foramen of Bochdalek was revealed behind it. The chest contained the third and fourth parts of the duodenum, all the small intestine, and all of the large intestine as far as the midtransverse colon. The structures were reduced, and the opening closed with mattress sutures of black silk. Convalescence was uneventful.

*Follow-Up.*—Roentgenologic examination six years postoperative shows the lung

FIG. 1.—Case 1: Hernia through the left foramen of Bochdalek.
Before operation: Small intestine in the left chest. Heart displaced to the right. Left lung collapsed.

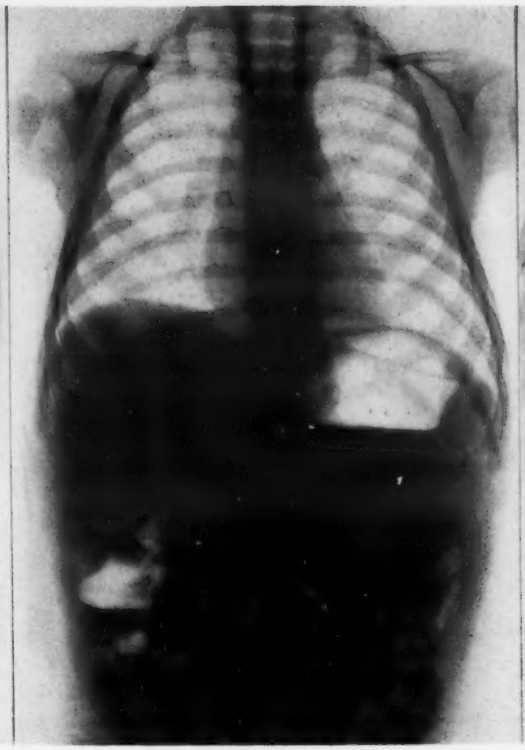

FIG. 2.—Case 1: After operation: Left lung expanded. Heart in normal position.

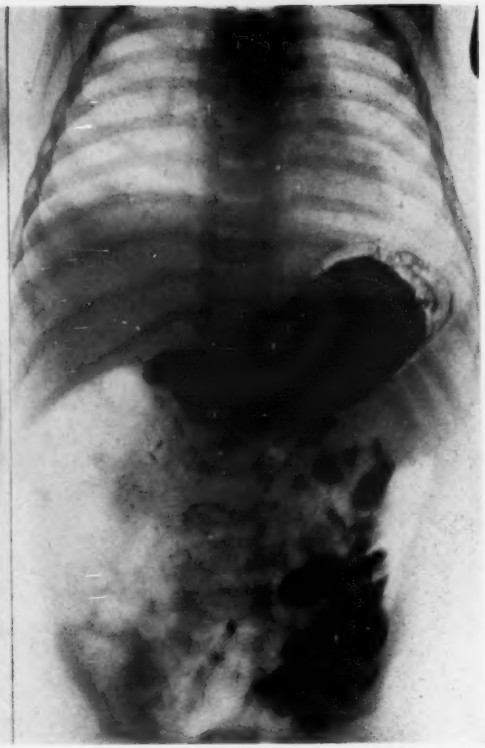

FIG. 3.—Case 1: After operation: All abdominal structures below the diaphragm.

FIG. 4.—Case 2: Hernia through the right foramen of Bochdalek. Before operation: Small intestine in the right chest. Heart displaced to the left. Right lung collapsed.

FIG. 5.—Case 2: After operation: Heart in normal position. Right lung expanded.

FIG. 6.—Case 2: All abdominal structures below the diaphragm.

FIG. 7.—Case 3: Hernia through the right foramen of Bochdalek.
Before operation: Colon in right chest. Heart displaced to the left. Right lung collapsed.

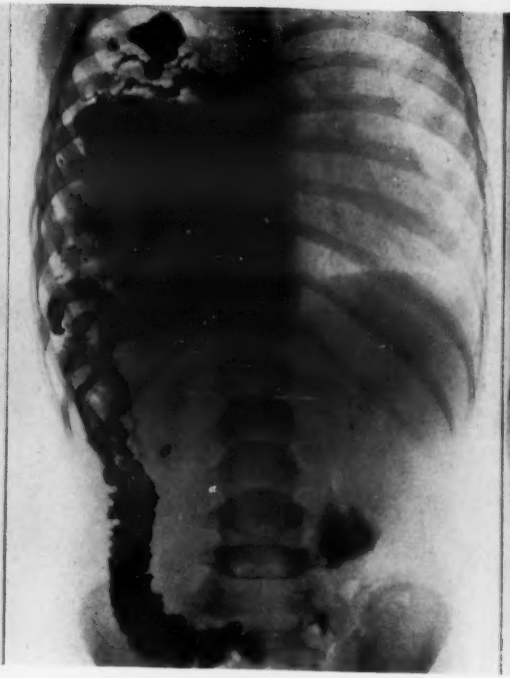

FIG. 8.—Case 3: Recurrence one and one-half years later: Heart displaced to the left. Right lung collapsed. Small intestine in the right chest.

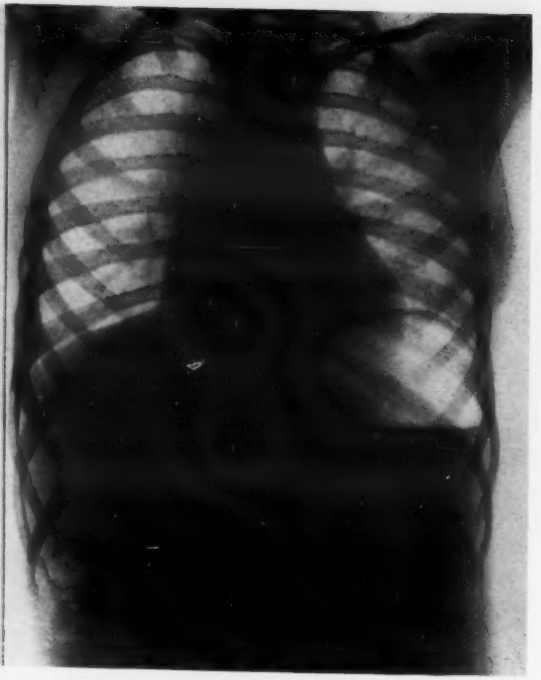

FIG. 9.—Case 3: One year after repair of recurrence: Heart in normal position. Right lung expanded. Abdominal structures below the diaphragm.

expanded, and all abdominal structures below the diaphragm. The child has been very well.

**Case 3.**—A. E., male, age 5 months, was admitted to the Babies Hospital, March 13, 1934, with a history of dyspnea and cyanosis since birth. Roentgenologic examination showed small and large intestine herniated into the chest through a defect in the right diaphragm.

*Operation.*—The abdomen was opened through a right subcostal incision, and disclosed a defect in the posterior half of the right diaphragm, which corresponded to about one-half the size of the entire right leaf. Closure seemed impossible. The structures in the chest consisted of: All of the small intestine beyond the duodenojejunal junction; the cecum; the appendix and right half of the transverse colon; and the right kidney. All viscera were, however, reduced, and the defect closed with considerable difficulty. Convalescence was uneventful.

The patient remained free from symptoms for about one and one-half years, when a routine roentgenogram of his chest, October 29, 1935, showed a recurrence.

*Second Operation.*—Age 2½ years: Twenty-four hours before repair, under avertin anesthesia, the right phrenic nerve was exposed in the neck, a loop of plain gut placed about it for traction, so that it could be readily identified and crushed if necessary. Under ether anesthesia, 24 hours later, the scar of the right subcostal incision of the previous operation was excised, and the abdomen opened. Practically all of the structures that were in the chest at the time of the first operation had reherniated into it. There were many adhesions present, and reduction of contents necessitated considerable dissection. In order to close the defect, a right intercostal incision was made through the eighth space, which greatly facilitated the closure of the diaphragmatic defect. The phrenic nerve was then crushed from within the chest. Closure was accomplished with mattress sutures of black silk. The patient developed pneumonia and pleural effusion in the right chest postoperatively; 275 cc. of sterile fluid were removed from the right chest upon two occasions.

FIG. 10.—Case 4: Hernia through the right foramen of Bochdalek.

Small intestine in the right chest. Heart displaced to the left. Right lung collapsed. Marked distention of the stomach and duodenum by gas is also to be seen, extending to the first segment of the jejunum; from which one may infer the existence of an obstruction in this location; and which condition was substantiated at operation.

*Follow-Up.*—It is now one year since the second operation, and roentgenologic examination, April 13, 1937, shows that the right lung is completely expanded; the heart is in normal position; and all of the abdominal viscera are below the diaphragm.

**Case 4.**—M. B., male, age 5 weeks, was admitted to the Babies Hospital, January 6, 1936, with a history of vomiting and cyanotic episodes for two days. "Was developing well until two days ago when he collapsed, turned blue and had rapid respirations." Had a similar spell 24 hours before admission. He had been delivered by cesarean section. Roentgenologic examination showed the right chest filled with small intestine. While under observation, the patient developed a high intestinal obstruction and was immediately operated upon.

After isolation of the phrenic nerve in the neck, a right subcostal incision was made. A large, posterolateral defect was found in the diaphragm through which practically all of the small intestine had herniated into the chest. A definite obstruction, due to a kink, was found in the upper jejunum. The opening in the diaphragm was closed with mattress sutures of black silk. The patient was placed in an oxygen tent immediately after operation, but he died 12 hours later as the result of shock.

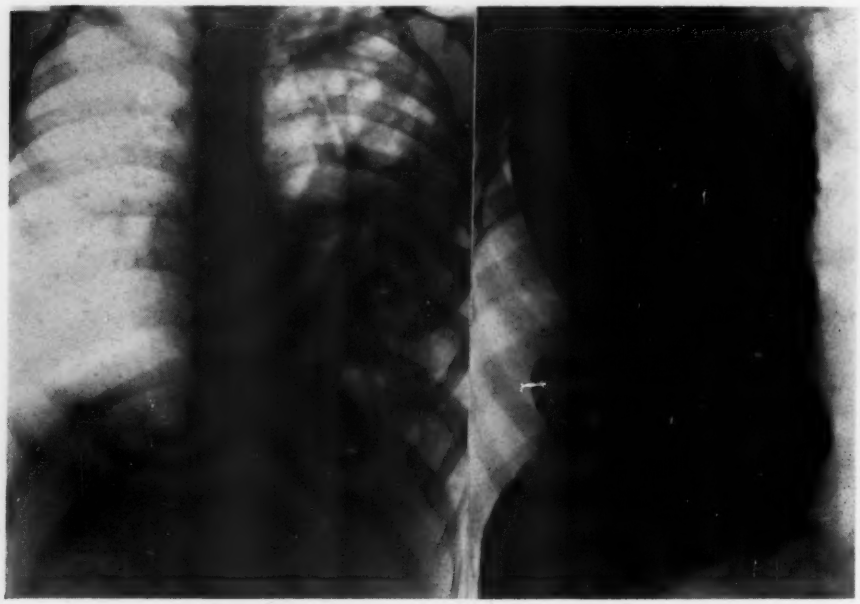

FIG. 11.—Case 5: Hernia through the left foramen of Bochdalek, and a defect in left dome of the diaphragm.

Gas bubbles in the left chest. Left lung is collapsed.

FIG. 12.—Case 5: Colon in the left chest, and very greatly dilated.

There was no choice but to operate upon this case. A palliative operation could not be performed because of the high intestinal obstruction. This is a serious complication, and one which may happen in any case where there is either small intestine or colon in the chest. This patient also had an umbilical hernia, bilateral cryptorchidism, penile hypospadias, hypertelorism, and a pilonidal sinus.

**Case 5.**—F. C., male, age 10 months, was first admitted to the Babies Hospital, in 1924, with the history of vomiting and convulsions. Roentgenologic examination showed the whole left chest filled with small intestine. He was discharged but was readmitted several times subsequently, with signs of an intestinal obstruction. Each time he was taken home against advice, when his obstructive symptoms had disappeared. He was finally admitted, when eight years old, with obstructive symptoms, at which time his parents consented to an operation.

FIG. 13.—Case 6: Hernia through the esophageal hiatus. Before operation: Stomach is in the chest. Esophagus is short. Transverse colon also found in the chest at operation.

FIG. 14.—Case 6: Three months after operation. Small part of the stomach has reherniated into the chest. Esophagus appears to be short.

FIG. 15.—Case 6: Transverse colon is entirely below the diaphragm.

FIG. 16.—Case 6: Lateral view showing appearance of colon with contrast media.

*Operation.*—April 25, 1932: The abdomen was opened through a left subcostal incision, which revealed a patent, left foramen of Bochdalek as well as a defect in the left dome. The left chest contained all of the small intestine from the duodenojejunal juncture on, all of the colon to a point beyond the splenic flexure, and the spleen. The structures were removed from the chest after a very tedious dissection. Both openings in the diaphragm were closed with mattress sutures of silk. The structures could not be returned to the abdomen because they had never been there before, and the cavity was not large enough to contain them. The child's condition was desperate in spite of a transfusion and an infusion on the operating table. His condition was so precarious, that the defects in the diaphragm were reopened and the viscera replaced into the chest. The patient died three hours postoperative. He should have been operated upon earlier in life, as his abdomen was not sufficiently developed to contain the viscera removed from his chest; in addition he should have had the phrenic nerve crushed before operation.

FIG. 17.—Case 7: Hernia through the esophageal hiatus.
No operation. Stomach in the chest.

FIG. 18.—Case 8: Hernia through the esophageal hiatus.
No operation. A large part of the stomach had herniated into the chest through the esophageal hiatus.

**Case 6.**—L. Y., female, age 6 months, was admitted to the Babies Hospital, June 22, 1931, with a history of loss of weight and vomiting for three months. She was a pale, undernourished child, whose roentgenologic examination showed her stomach and transverse colon herniated into her chest through the esophageal hiatus. She continued to vomit while under observation for two weeks. The vomitus contained blood on several occasions.

*Operation.*—July 8, 1931: The abdomen was opened through a right subcostal in-

cision. The entire stomach, the first and second portions of the duodenum, and almost all of the transverse colon were found to be in the chest, having herniated through a large esophageal hiatus. The viscera were reduced, and the esophageal hiatus was closed tightly around the esophagus after a large stomach tube had been passed. It was evident at operation that the esophagus was short, as it was only with considerable traction on the stomach that it could be kept in the abdomen. Convalescence was uneventful.

Roentgenologic examination, three months later, showed that a small portion of the stomach had reherniated into the chest. The patient has not been examined during the past year, but was doing well when last heard from. This was about the result we

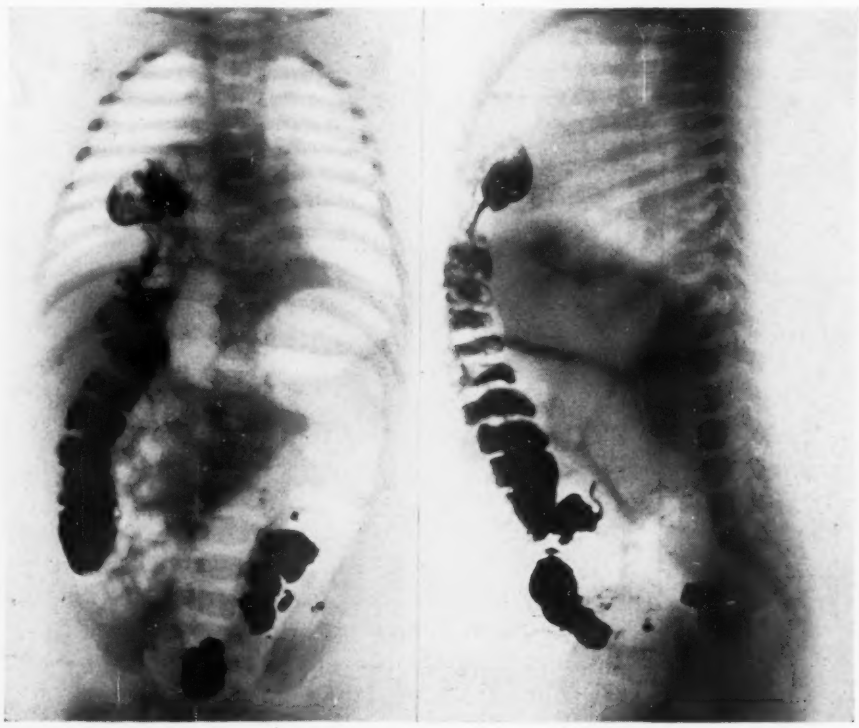

FIG. 19.—Case 9: Hernia through the foramen of Morgagni.
Part of the transverse colon in the chest, having herniated through the right foramen of Morgagni.

FIG. 20.—Case 9: Showing how helpful a lateral view may be in demonstrating the location of the defect in the diaphragm.

expected in this case. At least the operation kept the transverse colon from reentering the chest.

**Case 7.**—S. N., female, age 3½ years, was admitted to the Babies Hospital, November 16, 1932, with the history of paleness, and refusal of food and vomiting. The child had a waxy pallor, pale mucous membranes, and a systolic murmur at the apex. While under observation, she had a large hematemesis, and was transfused immediately. Her condition improved rapidly, but her mother refused to have the child operated upon, and took her home against advice.

Roentgenologic examination showed the stomach above the diaphragm.

Follow-up disclosed that the patient died of pneumonia during the past year.

**Case 8.**—D. S., age 2 months, was admitted to the Babies Hospital, October 10, 1933, suffering chiefly from malnutrition. This child also had a complete cleft palate.

Roentgenologic examination showed the stomach in the chest, having herniated through the esophageal hiatus. The patient was taken from the hospital against advice, without operation. Follow-up report, one month ago, stated that the child was doing very well.

**Case 9.**—J. H., male, age 3 months, was admitted to the Babies Hospital, April 1, 1932, with a history of cough and fever for the past two weeks. The child was pale, poorly developed, poorly nourished, and typical in every way of a Mongolian idiot. Roentgenologic examination showed bilateral bronchopneumonia. A later gastro-intestinal series showed a large part of the colon to have herniated into the right chest through the foramen of Morgagni. He developed diarrhea and intestinal hemorrhage, and died without operation.

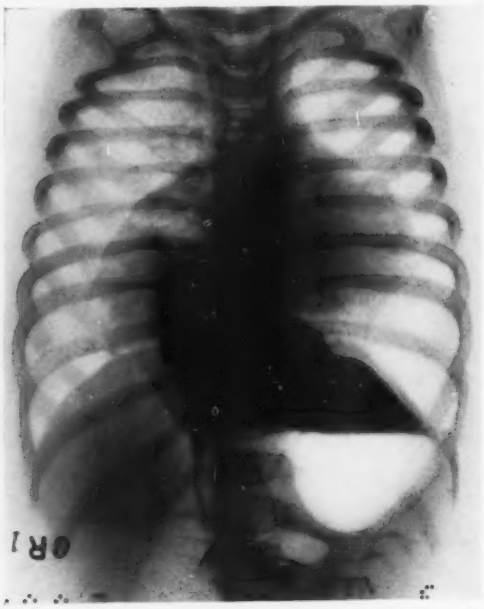

FIG. 21.—Case 10: Hernia through the foramen of Morgagni.
Part of the colon in the chest to the right of the sternum.

**Case 10.**—R. D., age 11 months, was admitted to the Babies Hospital, February 3, 1937, with a history of shortness of breath since the age of two months. The patient had always been a feeding problem, and was a typical Mongolian idiot. Roentgenologic examination showed a herniation of the colon through the right foramen of Morgagni. There was a definite upper respiratory infection present, and the child was discharged because of this, but is to return for operation.

*Results.*—In this group of cases, seven operations were performed upon six of the ten patients. One case is to be operated upon when he recovers from a respiratory infection. Two patients died from shock, one, a five weeks old infant who developed a high intestinal obstruction while being prepared for operation, and the other, a boy, age 8, died after a long and tedious dissection of structures adherent in the chest and the closure of two diaphragmatic defects. One case of esophageal hiatus hernia has a partial recurrence due to a short esophagus, but he is doing well. One patient operated upon

at the age of five months had a recurrence one and one-half years later. The operation for recurrence seems successful, two years later. One case with hernia through the left foramen of Bochdalek is well seven years after operation. One case with hernia through the right foramen of Bochdalek is well six years after operation.

Five of these ten patients showed no congenital abnormalities other than the diaphragmatic hernia. Both patients with herniae through the foramen of Morgagni were Mongolian idiots. One of these also had a congenital heart derangement. One case of esophageal hiatus hernia had a cleft palate. One patient (Case 4) also had an umbilical hernia, bilateral cryptorchidism, penile hypospadias, hypertelorism and a pilonidal sinus.

## SUMMARY

(1) The symptons of diaphragmatic hernia may be either respiratory or gastro-intestinal in origin, depending upon what viscera are contained in the hernia and what structures in the chest are pressed upon by the hernial contents.

(2) Many cases of diaphragmatic hernia are symptom-free and are discovered in the course of a routine roentgenologic examination.

(3) The diagnosis of diaphragmatic hernia is most surely arrived at by a roentgenologic study.

(4) Treatment should be surgical repair of the hernia in all cases where any portion of the intestinal tract is involved, because of the danger of intestinal obstruction.

(5) The author prefers the abdominal approach, but there are many surgeons who prefer the thoracic approach.

(6) Positive pressure anesthesia is very desirable.

(7) Patients should be placed in oxygen tents after operation.

(8) Pleural effusion may occur after operation, and should be aspirated.

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# DIRECT INGUINAL HERNIA*

## PRESENTATION OF AN OPERATION FOR ITS CURE

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HALSTED[14] states that an operation for the cure of inguinal hernia was devised and published by Heliodorus who lived during the time of the Roman Empire. This operation corresponded to some extent to operations performed during our time.

*The Bassini Operation.*—It was not, however, until Bassini[5, 23] devised his operation, in 1884, that a procedure was presented which was based upon exact anatomic studies. The earliest description of this operation that I have been able to find was that published, in 1890, in the Archiv. für klinische chirurgie, Berlin. In this he referred to the work of Wood, von Segond, von Lucas-Championniere and Czerny, all of whom had devised operations for the cure of hernia. He states that Julliard had only a single radical cure in 22 operations. It seems that following all of these operations the patient needed to wear a truss to prevent recurrence. He himself first operated by the Czerny method, in which the sac was dissected out and cut away after tying its neck with catgut. The neck was then thrust into the abdominal cavity and the wound closed with catgut sutures. He undertook to improve results by devising first an operation which was similar to the one devised by Macewen about the same time, in which after ligating and excising the sac, the neck was sutured under the aponeurosis of the external oblique, and the process of the neck beneath the ligation was gathered up with sutures to form a pad which was supposed to be a bulwark against any further descent of hernia. Subsequent investigations showed him that this pad was quickly absorbed, and his operation was no better than that of his predecessors. He then devised the operation in 1884 which has since borne his name. After having described the operation he states the principle of it as follows: "In both cases, that is in external (acquired or congenital), or in internal (direct) herniae, the inguinal canal is reconstructed after the manner of its physiologic formation, that is, with an abdominal and subcutaneous opening, of which the first lies external to the second; and is provided with two walls, one posterior and the other anterior. Under the influence of abdominal pressure, the new posterior wall is pressed against the anterior and both support each other in order to withstand the continuous and strong impulse of the abdominal viscera, while the spermatic cord is permitted to pass through between them. The new abdominal opening and the new posterior wall of the inguinal canal are formed from muscular and aponeurotic tissues which functionate and therefore cannot disappear by resorption."

*Recurrences.*—It is interesting to note that in our country the failure to

* Read before the Southern Surgical Association, Birmingham, Ala., December 9, 1937.

secure a radical cure by operations in vogue previous to the Bassini type is set forth in a statement by Dr. W. T. Bull, as quoted by Halsted: "Now [1890] that ten years have elapsed since the modern radical operations have been in vogue, we ought to hear of, or have presented to us, patients who have been more than five years at the least, without relapse. We could naturally expect to see such cases occasionally, but there are none such."

*Other Operative Procedures.*—Wm. S. Halsted devised his operation in 1888,[16] and first published his results, as far as my researches go, in 1893. Alexander H. Ferguson,[10] in his first paper published in 1895, reported an operation in which he followed the technic of Macewen in plicating the sac to the inner surface of the abdominal wall to block another descent of the hernia. The cord was placed external to the aponeurosis of the external oblique. His second operation,[11] and the one that bears his name, was published in 1899.

Without contrasting the merits of these three operations, Bassini, Halsted and Ferguson, they are all sufficiently alike to suggest a common origin. They all provide for the complete removal of the sac and the reconstruction and reinforcement of the abdominal wall. Their chief difference is in the method of dealing with the cord. It would appear that the suggestion for the methods of Halsted and Ferguson was from Bassini's operation. There can be no question of the value of these methods, and the large number of cases permanently cured by them is a sufficient testimony in their behalf.

However, there were more or less numerous recurrences reported by various surgeons, and the reasons for this, and the various methods suggested to overcome recurrence, have given rise to a voluminous mass of surgical literature. It seems to be generally conceded now that where there is recurrence after the neck had been properly ligated and the sac removed, it occurs below the abdominal opening of the canal.

*Operations Designed to Prevent Recurrence.*—The following are some of the principal methods adopted to prevent recurrence.

In 1901, L. L. McArthur[19] reported his operation for strengthening the suture line between the muscles and conjoined tendon and Poupart's ligament by the use of strips of fascia derived from the pillars of the external ring and extended up to the belly of the external oblique muscle. In 1904, he[20] reported again on the same operation.

In 1906, E. W. Andrews[3] reported on imbrication of the aponeurosis of the external oblique.

In 1920, Hoguet[15] reported on folding back the medial flap of the aponeurosis and including that in the sutures of the muscle to Poupart's ligament, after which the edge of this flap was sutured to the lateral flap over the cord.

In 1924, Edmund Andrews[1, 2] reported on exposing the thickened part of the transversalis fascia and suturing that to Poupart's ligament.

There have been many other procedures advocated, but those mentioned above express the general ideas.

*Other Explanations of Recurrence.*—Another explanation of recurrences was that offered by Seelig and Chouke,[25] in 1923. They made the contention that muscle sutured to fascia did not unite with it, but that what took place was simply an adhesion which becomes attenuated under pressure. Still another explanation was that suture of muscle was harmful.

In 1923, Sir Arthur Keith[17] described the shutter action of the muscles surrounding the inguinal canal by which these muscles contract and close the canal to counteract pressure from above.

In 1925, R. Hamilton Russell[24] advocated proper removal of the sac alone in the cure of simple indirect hernia. This had been his practice in operating at a children's hospital and he had extended the operation to include younger adults before deformity and distortion had taken place. He was of the opinion that suture of the muscle injured it and prevented its action and predisposed to recurrence.

In 1933, J. D. Rives[22] expressed the same opinion and gave three cardinal principles as follows: (1) Muscle deprived of function atrophies; (2) muscle offers no considerable resistance to forces directed perpendicular to its fibers; (3) fascia must be applied directly over the defect. For this purpose he advocated the Andrews imbrication.

All of these have a basis of sound reasoning, and of all the modifications, Seelig's (1927) method[26] of applying the McArthur fascial suture appears to me to be the best. However, no operation that depends for its permanency upon the suture of muscle or fascia to Poupart's ligament will be uniformly successful in the cure of all types of inguinal hernia.

*The Chief Reason for Recurrence.*—This weakness consists of the absence of a well-formed conjoined tendon and attenuation of the muscles that form it. It is to this point that I wish particularly to draw attention.

The difficulties encountered in effecting a cure of direct hernia by the methods referred to, the reasons for recurrence, and the presentation of an operative procedure, which in my hands, thus far, has effected cures in 100 per cent of 27 operations, is the object of this paper. That the methods commonly in use have a high percentage of recurrence is shown by many published reports, of which the statistics given in Christopher's Surgery,[7] published in 1936 (page 1376), are a fair sample. The section on hernia is written by Seward Erdman and he states: "From a number of the largest and best surgical clinics, follow-up reports based on actual examinations show from three to seven per cent recurrence after operations for indirect hernia; and from 15 to 30 per cent after operations for direct hernia."

That there is a defect at the point where direct hernia occurs was early discovered by Bassini, but the methods that he advocated for overcoming this were ineffectual. Halsted, and Bloodgood[6] endeavored to fill in this defect by suturing the rectus muscle or a flap from its fascia to Poupart's ligament, and W. A. Downes[8] advocated the same procedure. However, in a later paper he[9] states: "The problem of inguinal hernia resolves itself into the

management of the direct variety. I have personally reached the conclusion that a certain number of direct herniae cannot be cured by operation."

Joseph C. Bloodgood states: "The chief cause of recurrence in the lower angle of the wound, stated in a previous report of operations at Johns Hopkins Hospital, in 1899, was due to the fact that, whether the hernia was direct or indirect, the conjoined tendon was weak or obliterated, and the ordinary suture or closure of the defect in the abdominal wall was not sufficiently strong to protect from recurrence in the lower angle, and that the transplantation of the rectus muscle and its fascia was not a certain cure."

*Diagnosis of Direct Hernia.*—In diagnosing direct hernia, the scrotum is invaginated by the examining finger which passes directly over the pubic bone into the pelvic cavity. Sometimes two fingers can be introduced and swept freely around the inner walls of the pelvis and beyond the rectus muscle, which is felt to the medial side of a triangle, the base of which is formed by the pubic bone, and the lateral sides by Poupart's ligament and the internal oblique and transversalis muscles attached to Poupart's ligament. If there is no sac present descending from the abdominal ring, it must be obvious that constructing a new canal for the cord, whatever the method employed, will have very little bearing on the closure of this defect.

*Deficient Structures at the Pubic End of Canal.*—Dissection of the tissues involved will show that the conjoined tendon is absent and that the internal oblique and transversalis muscles pass over directly to the rectus sheath, leaving a defect which is often as much as one and one-half inches from the lower border of the muscle to the pubic bone and Poupart's ligament. In addition the fibers of the internal oblique muscle at the lower border are attenuated and weak. The greatest defect is at the border of the rectus and from there it extends lateralward with a diminishing width, often as far as the abdominal ring. In this way the floor of the canal is often weak throughout its extent.

Attempts to close this defect by the ordinary methods of suture are not uniformly successful. Sutures in vogue affect only adhesions if successful, but there is a distinct gap, and efforts to close this require that the suture be applied under tension. This is a violation of the cardinal principles of surgery. The sutures either cut out or absorb and the gap recurs. Neither can the fascia of the external oblique be overlapped here because the pillars of the ring at this point are fixed by their insertion into the pubes. In addition to that, the base of the triangle that is formed by suturing the internal flat muscles to Poupart's ligament is the pubic bone to which nothing is attached, consequently the angle cannot be completely obliterated.

*Normal Structures at Pubic End of Canal.*—Normally the opening of the subcutaneous ring is protected by the conjoined tendon, which is inserted into the crest of the pubes and along the pectineal line behind the attachment of Gimbernat's or the lacunar ligament for the distance of about 1.25 cm. The pectineal line is covered by a strong band of fascia called Cooper's ligament, which extends lateralward from the pubic tubercule and merges

with the pectineal fascia. It is to this that Gimbernat's ligament and the conjoined tendon are attached.

The transversalis fascia in Hesselbach's triangle, especially in cases of direct hernia, is thin and attenuated and can offer very little resistance unless supported by other tissues. The principal support is derived from the conjoined tendon.

*Essentials of the Proposed Operation.*—Before proceeding to describe the operation, it would be well to direct attention to two elements that are essential parts of the operation about to be described: namely, fascial suture and the fascial or ligamentous covering of the superior surface of the pubic bone.

The opening of the superficial inguinal ring is situated anteriorly, and immediately over the insertion of the conjoined tendon. Absence or attenuation of the conjoined tendon consequently creates a definite weakness at this point and is undoubtedly the cause of most instances of direct hernia.

*The Use of the Fascial Suture.*—The use of fascia in operations for hernia was first reported by L. L. McArthur,[19] in 1901, who published a second article[20] on the subject, in 1904. His method was employed in conjunction with the Bassini operation. In it he made use of strips of fascia derived from the aponeurosis of the external oblique. He states that other methods of suture are dependent for success upon the formation of cicatricial tissue between the opposing surfaces, and that we must expect occasional failure because of this very yielding cicatricial tissue, but that he had been able to demonstrate that his autoplastic suture heals in situ, is not absorbed and does not slough. He presented microscopic specimens in verification of his statements.

Gallie and Le Mesurier,[12, 13] in 1921 and 1923, called attention to the advantages of fascial suture. They proved beyond any doubt that fascia continues to live in an unchanged form after being used as suture material. It is, therefore, to be regarded as a transplant and not merely as a suture. In their cases the fascia was derived from the thigh, and they advised that it be woven over the defect. Case 12, upon whom an autopsy was performed, 57 days postoperative, illustrates the permanence of the fascial suture. Microscopic examination demonstrated that the histologic character of the fascial transplant had remained unchanged and that it was firmly anchored in the tissues through which it passed (Figs. 1, 2 and 3).

It is somewhat difficult to understand why the definite advantage resulting from the employment of fascia in the cure of hernia has not been more generally adopted. At about the time that McArthur presented his operation and advocated the use of fascial strips, there began to appear on the market various types of suture material, plain and chromic catgut, kangaroo tendon, silk, *etc.*, that were put up and sterilized in glass containers. As they were ready for immediate use, and were easily obtainable and convenient, they came into general use for hernia as well as for other types of surgery.

There is, however, an essential difference between fascia and all other

PHOTOMICROGRAPHS OF THE FASCIAL SUTURE REMOVED FROM CASE 12, WHO DIED 57 DAYS POSTOPERATIVE, FROM THROMBOSIS OF THE LEFT COMMON ILIAC ARTERY AND VENA CAVA.

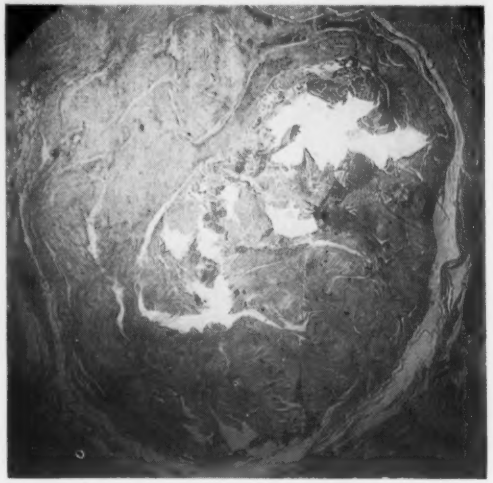


FIG. 1.—Photomicrograph showing cross-section of the fascial suture. The clear irregular areas in the center are artifacts, due to tearing of the tissues in sectioning. The clear area surrounding the fascial suture is also an artifact, due either to pulling on the stitch before it was imbedded or to an uneven shrinking of the tissues in the fixing solution. The denser character of the fascial stitch can be made out even at this magnification. At the upper left of the suture is adipose tissue. At the lower left are a group of blood vessels in the connective tissue surrounding the suture. (Low power.)

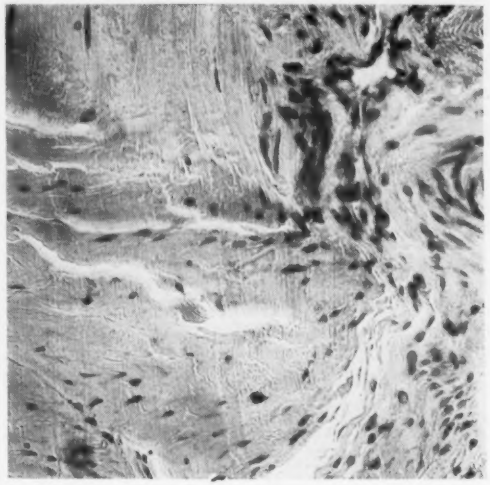


FIG. 2.—Photomicrograph of area shown in Figure 1. On the left can be seen the dense, compact fibrous structure of the fascial stitch. A few scattered nuclei attest the viability of the tissue. To the right is the looser connective tissue through which the stitch passes. The large, pale vesicular nuclei indicate young, growing fibroblasts. In the upper center is a small blood vessel and just below this, what appears to be new forming capillaries. (High power.)

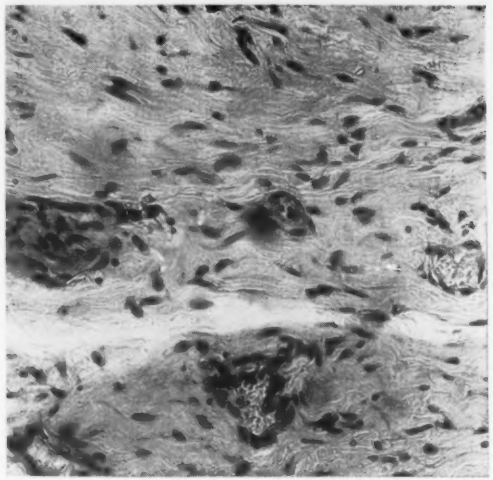


FIG. 3.—Photomicrograph of an area on the edge of the fascial suture (top). This shows the many new capillaries forming in the areolar tissue at the edge of the suture. Blood cells can be seen in both of these. (High power.)

sutures. Fascia is a living transplant which remains permanently where it is placed, while all other forms of suture are eventually absorbed or cut out, and the strength of the wound is dependent upon the union that takes place while absorption is going on. This difference appears to have been overlooked by the profession.

The difficulties of securing fascia from the thigh as advocated by Gallie and Le Mesurier, and the complicated weaving necessitated, together with the additional time consumed, have been a bar to the employment of their method. But the fact that recurrence of herniae during the war was so great as to disqualify enlistment of men who either had hernia or had been operated upon for it called attention forcibly to the necessity of devising some procedure that would make the cure of hernia more permanent.

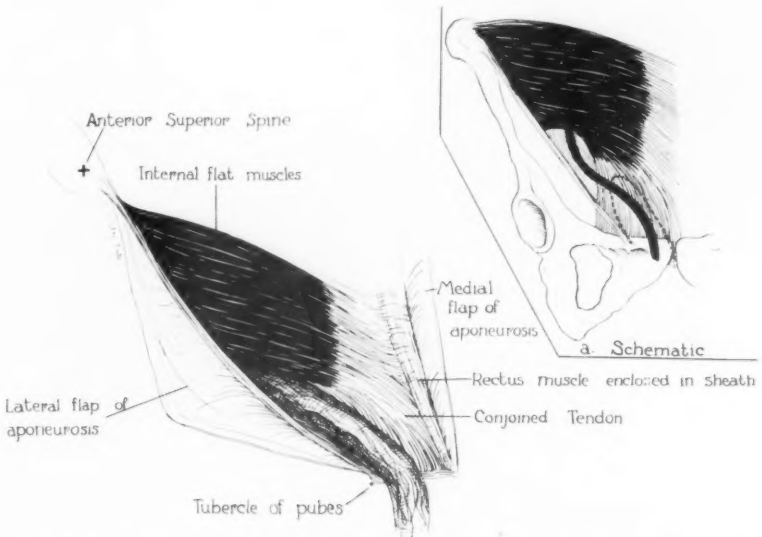
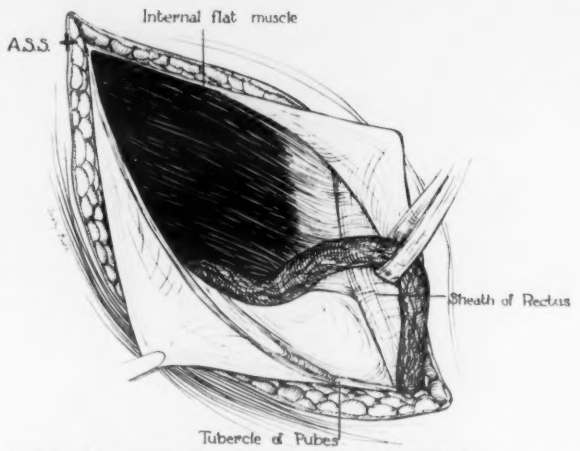



FIG. 4.—Normal deep layer of muscles. (Redrawn from Cunningham's Manual.) The deep flat muscles have their origin from the internal half of Poupart's ligament and lie anterior to the abdominal ring. That portion forming the conjoined tendon is inserted behind the cord and the external inguinal ring, the location of which is indicated by the dotted line.

Referring to the fascial covering of the pubic bone, the first suggestion as to its employment in operating for the radical cure of hernia, as far as can be ascertained, was made by W. Wayne Babcock,[4] in 1927. The fascia covering the superior surface of the pubic bone is several millimeters thick, and is composed of extensions on to the bone of Poupart's, Gimbernat's, Cooper's and the superior ligament of the symphysis. It is tough, firmly fixed to the bone and is, therefore, admirably adapted for suture. It is of particular value because the bone at this point forms the base of the triangle, already referred to, that cannot be successfully obliterated in any other manner except by direct suture to the bone at this point. The operation described by Babcock was an ingenious one and was performed with a single suture of chromic catgut. It was because of a recurrence following this operation, in which chromic cat-




(Drawn from operated case by actual measurement.)

FIG. 5.—In direct hernia the lower fibers of the flat muscles are deficient, so that, as a rule, there is no conjoined tendon, and the fibers of the muscle pass directly over the lower border.

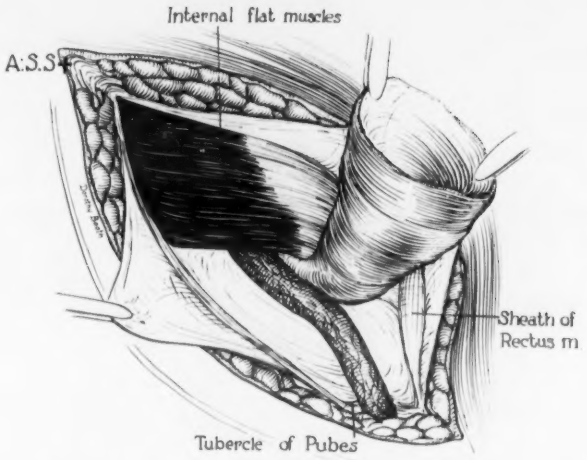



FIG. 6.—The defect on the lower border of the flat muscle makes a weak bed for the inguinal canal, which undoubtedly favors the development of indirect hernia, and favors recurrence of indirect hernia after operation, as well as being the cause of direct hernia. In this case there were both an indirect hernia and a direct hernia. (Drawn from operated case by actual measurement.)

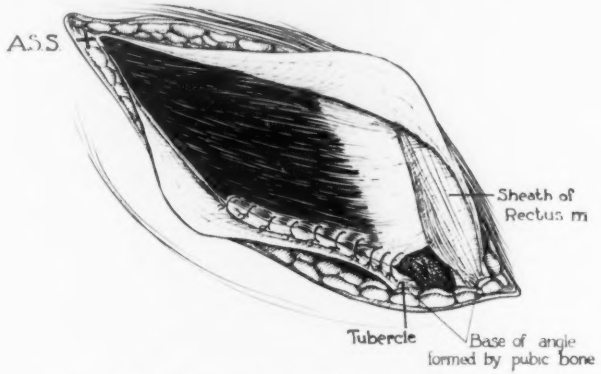



FIG. 7.—Angle that remains when tissues are sutured from side-to-side. Note that the bone which forms the base of the angle is uncovered and, therefore, the defect is not obliterated. (Drawn from operated case by actual measurement.)

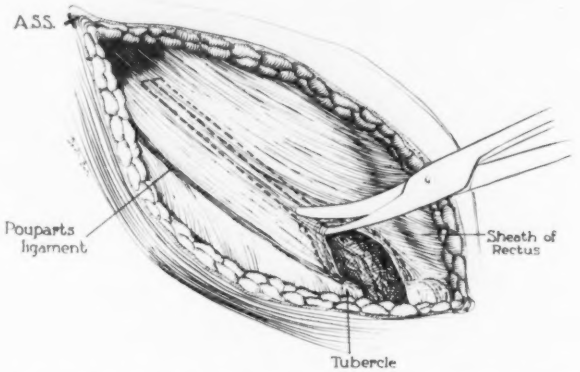



FIG. 8.—Outlining the fascial sutures. Incisions should all commence at the pubes in order to preserve a firm attachment.

gut was employed, that I was led to investigate, more completely, the subject of the radical cure of hernia.

It was to overcome the various weaknesses that appeared to have been present in other operations that the operation herewith described was devised. Free use has been made of the studies, investigations and work of others, which is hereby acknowledged.

*Operative Technic.*—(1) The incision is made down to the aponeurosis of the external oblique and in the direction of its fibers from a point sufficiently high to expose the belly of this muscle and extending down over the pubes sufficiently low to permit adequate exposure of the structures about the external ring. The flaps are carried back on either side to give wide and clean exposure of the aponeurosis.

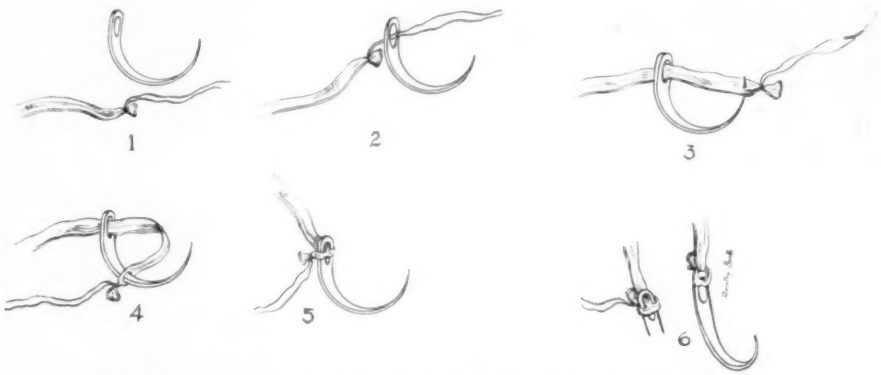



FIG. 9.—Illustrating the method of threading the fascial suture on the needle.

(2) The external spermatic fascia is dissected from the borders of the external ring and the pillars, and their origin exposed and isolated well down over the pubic bone, care being taken to free them of connective tissue. The canal is then opened by incising the aponeurosis from a point at the center of the apex of the external ring to the belly of the muscle, care being taken to follow the fibers of the aponeurosis and not to cut across them. This is accomplished with a pair of scissors which are only partly opened and are pushed along without a cutting motion. The two flaps of the aponeurosis are then separated from the underlying structures, both lateral- and medialward, exposing Poupart's ligament, particularly around the pubic tubercle to which it is attached, and then the internal oblique well beyond the border of the sheath of the rectus and down over the pubes.

(3) The cord should then be carefully examined for any evidence of a sac of an indirect hernia. This is not infrequently found and if present it should be isolated, the neck twisted, ligated, and removed. The cord is then freed in its lower portion and the area of the direct hernia explored. As a rule the protrusion of the sac has disappeared unless it happens to be a very large one. I have never found that any good was accomplished by resecting these tissues in the presence of ordinary relaxation. If the sac is large it is,



angle is uncovered and, therefore, the defect is not obliterated.
(Drawn from operated case by actual measurement.)

of course, resected. It will usually be found that there is a wide space between the lower border of the internal flat muscles and Poupart's ligament and the pubic bone, and that the conjoined tendon is either absent or else very attenuated.

(4) In securing the fascial sutures, certain points are necessary in order to avoid unfortunate accidents. If the width of the strip is measured at the belly it will be found, when the pubic end is reached, that the strip may have been entirely severed by passing into the ring itself or have only a few inade-

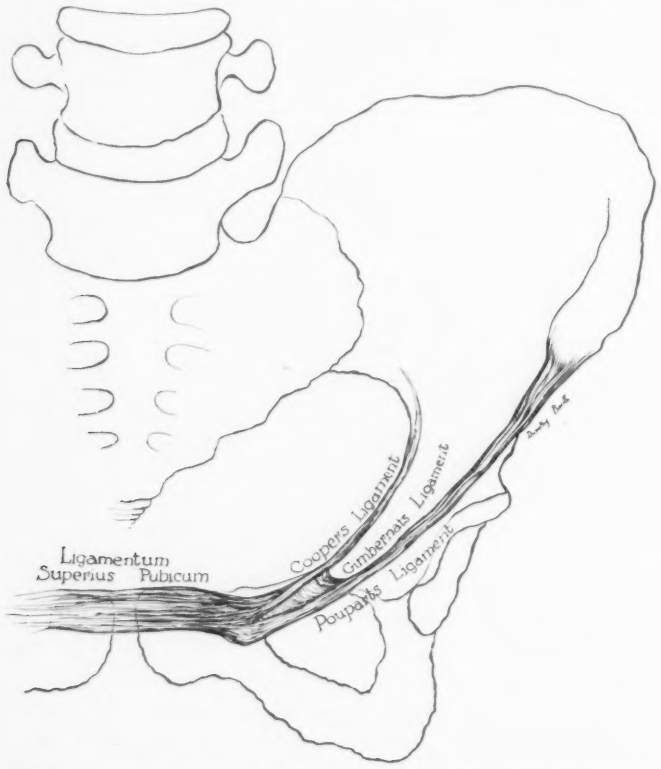



FIG. 10.—The thick, tough layer of fascia firmly attached to the superior surface of the pubic bone, which appears to be extensions on to the bone of Poupart's, Gimbernat's, Cooper's, and the superior pubic ligaments.

quate fibers remaining. As the firm attachment of the strip to the pubis is of prime necessity, the isolation of the strip should commence at the pubic end. After the initial incision, scissors may well be employed for completing the strip as already described. The medial strip is made first.

The next problem is threading the strip on the fascia carrying needle. If it is simply passed through the eye of the needle and then folded over and tied to the strip behind the eye, it will frequently slip out. The best method is to first tie the end securely to prevent its splitting, leaving the ends of the tie long enough to thread through the eye of the needle easily, bringing the end of the strip through from the posterior surface for a sufficient distance, so

that the point of the needle can pierce the end of the strip behind the ligature. The pierced end of the strip is then slipped back over the eye of the needle, which makes a friction knot that holds securely and uses up very little of the fascia.

The fixation of the end of the suture is important. Operators who have used fascial sutures have various ways of fixing the terminal knot. I do not use the knot for two reasons. One is that the knot uses up fascia unnecessarily which at best is not too abundant. The other is that the simple method of fixing the end to underlying fascia or to the suture itself by suture is better. This is best effected with fine silk. The stitch commences with a bite in the underlying fascia or suture and comes up through the edge of the suture and is carried back on the other edge in the same manner, after which it is tied. Two of these sutures are used.

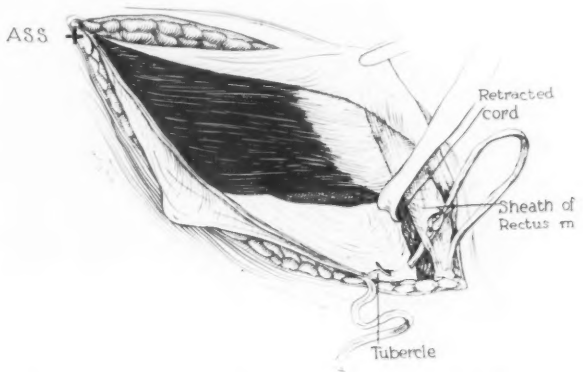



FIG. 11.—Tissues to be sutured, showing first stitch being introduced. This fascial suture is derived from the medial pillar and is carried through the rectus sheath and muscle and then passes over the cord, and takes a firm bite in the fascia covering the pubic bone.

(5) The cord is then held back medialward under the rectus by a small retractor and the suture is passed through the rectus sheath and the muscle and over the cord and then takes a good bite in the fascia that covers the pubic bone at the pecten ossis pubis.

This last point is to be selected so that the suture may not unduly compress the cord and at the same time fit it very snugly. This forms a new ring for the cord that is strong and unyielding, because the fascia passes over the cord in the direction of its fibers. It is not like the normal external ring, in which the application of force separates the fibers. The fascial stitch is then continued, passing through the rectus in the same manner and is sutured to the pubic bone. As a rule only two stitches include the rectus and the pubic bone, the last being passed at a point near the tubercle. After that the suture embraces the thick part of the internal flat muscles together with the transversalis fascia and Poupart's ligament. In passing the fascial suture it must be borne in mind that it is not a suture for approximating

surfaces but is transplanted tissue, and that the object is to secure good anchorage; therefore, the thicker the tissue through which it passes, the better. When Poupart's ligament is reached the first stitch should embrace Gimbernat's ligament, after which it is continued on Poupart's ligament until the point of origin of the internal flat muscles is reached. It is usually

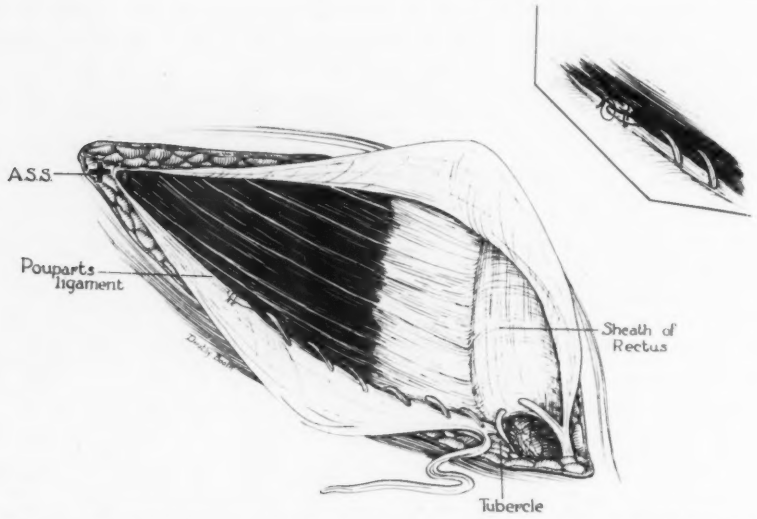



FIG. 12.—The first suture line completed. The suture passes over the cord and forms a firm ring. Care must be taken not to compress the cord unduly.

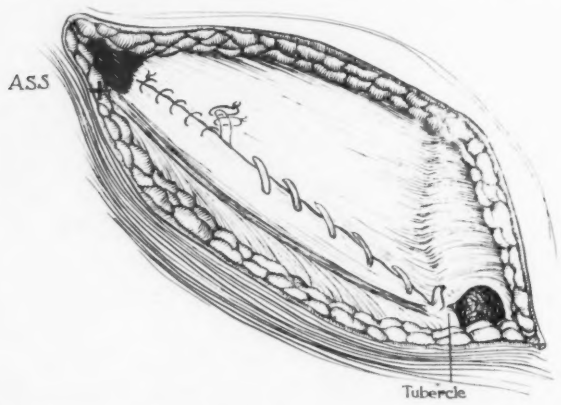



FIG. 13.—Second suture line completed. The suture passes from the pubic tubercle, to which it is firmly attached, to the opposite leaf of the fascia, making a new and undilatable external ring.

long enough for this purpose. If not, one or two stitches of chromic catgut may be employed to complete the line of suture. As there is little if any tension at this point, chromic catgut is sufficient.

(6) The second fascial suture is derived from the lateral pillar. After being prepared and fixed to the needle it is employed to close the interval left in the aponeurosis after the removal of the strips. The stitch is first

passed just over the emergence of the cord to the opposite edge of the aponeurosis and continued until the gap is closed. If not sufficient for this, the suture line may be concluded with chromic catgut, as there is no tension here and the suture line is immediately over the thick belly of the internal flat muscles.

It will be observed that by passing this last suture across to the opposite leaf of the aponeurosis, just above the cord, this point, which is normally the weak point, is further strengthened, because the fascia extends across in the direction of its fibers and makes a strong closure, whereas, if the fibers of the aponeurosis are simply sutured together, bringing their lateral aspects in juxtaposition, it is much easier for the fibers to be separated by continual pressure from above.

## COMMENT

Normally there is no muscle behind the cord except the conjoined tendon, and the canal is closed by the shutter action described. When there is a defect in the muscle impairing this action, it is necessary to carry the fascial suture up to close this defect. By taking a deep bite through the thick part of the muscle, the fascial strip comes into contact with the perimysium of the muscle and is firmly anchored. In addition, the suture acts as a tendinous attachment to the structures to which the muscle is sutured.

It is a mistake to employ only one procedure for all herniae. The operation should be predicated upon the existing pathology.

The surgical rules that apply to suturing under tension do not apply to the fascial suture. It does not cut out or absorb but becomes a permanent transplant.

When a sac of an indirect hernia is present it is removed by high ligation and covering the stump over by a purse string suture of the transversalis fascia. This opening lies behind the origin of the flat internal muscles which amply protect it.

The advantage of separating the various layers in the abdominal wall is not only to facilitate the operation but also to permit them to fall into a new relationship after the suture is completed, thereby affording additional support.

The fascia is resistant to infection. Where infection has occurred in dealing with very large herniae, especially when incarcerated, the fascia is not involved. I have seen the glistening fascia where it was exposed and it has remained viable and effective.

The operation described protects the external ring even more effectively than the normal insertion of the conjoined tendon does.

Every case of hernia should be examined for weakness at the pubic end and whenever found the defect should be closed as described, irrespective of what may have been done in order to repair any coexisting hernia.

The cord is allowed to remain without disturbing its attachments more than is necessary to remove the sac, should one be present.

When for any reason the fascia from the pillars of the ring is inadequate, fascia can be secured from the fascia lata in abundance by using the Masson[18] stripper.

The chief defect in earlier operations for inguinal hernia, as far as direct hernia is concerned, was in the attempt to close the angular defect by suture from side-to-side, which cannot be accomplished satisfactorily.

The introdution of the autogenous fascial suture has markedly improved results in the operation for inguinal hernia as a whole, as the results obtained by the Mayo Clinic, Seelig[26] and Payne[21] show. Still, it does not appear that the fascial suture has been as generally adopted as its value would seem to warrant. The high percentage of recurrences following operations for direct hernia has been an embarrassment to surgeons, which I believe the employment of the procedure herewith suggested will obviate. We must get away from viewing the sac as the exclusive cause of hernia and direct attention to the anatomic defect at the pubic end of the canal, which not only is the cause of direct hernia, but is, I believe, a contributing factor in the development of indirect hernia.

The operation herewith presented can be almost as easily and quickly performed as those in which prepared sutures are employed. The only additional time that is required is for the preparation and threading of the fascial sutures.

## CASE REPORTS

**Case 1.**—No. 39–159. June 21, 1934: Male, age 61, farmer.

*Duration.*—First noticed in 1917. Operated in 1925. Recurrence several weeks ago.

*Physical examination.*—Right direct inguinal hernia.

*Operative findings.*—Round aperture lying on pubic bone with irregular sac.

*Operative procedure.*—Sac isolated, ligated and excised. Operation completed as described.

*Result.*—April, 1938: Doctor reports no evidence of hernia.

**Case 2.**—No. 40–15. January 21, 1935: Male, age 27, laborer.

*Duration.*—Five years.

*Physical examination.*—Double hernia. On both sides, on invagination of scrotum, finger passes directly over pubic bone into pelvic cavity. On right side hernia comes down into scrotum.

*Operative findings.*—Indirect sac on both sides.

*Operative procedure.*—Sac isolated, ligated and excised, and reinforced by suturing transversalis fascia over neck. Operation completed as described.

*Results.*—April, 1938: Doctor reports no evidence of hernia.

**Case 3.**—No. 40–32. January 24, 1935: Male, age 19, clerk.

*Duration.*—Two years.

*Physical examination.*—Left inguinal hernia. Does not enter scrotum. On examination, finger passes over pubic bone into abdomen. Can palpate edge of rectus.

*Operative findings.—No sac* in relation to cord, lower segment internal flat muscles and conjoined tendon deficient. Fibers of muscle pass directly to rectus sheath.

*Operative procedure.*—Operation completed as described.

*Results.*—Has moved out of the state but reported over phone: "Has no trouble, perfect result." Since above note, patient returned to city for examination. No evidence of hernia was found.

**Case 4.**—No. 40–52. February 21, 1935: Male, age 48, bank official.

*Duration.*—First noticed 21 years ago.

*Physical examination.*—Both external rings enlarged and has protrusion at external rings. On invagination of scrotum, on both sides, finger passes over pubic bone directly into abdominal cavity.

*Operative findings.*—Deficiency of lower segment of internal flat muscles and conjoined tendon on both sides.

*Operative procedure.*—Operation on both sides completed as described.

*Result.*—April, 1938: Examined by myself. No evidence of hernia.

**Case 5.**—No. 40-64. March 8, 1935: Male, age 36, farmer.

*Physical examination.*—Both inguinal rings open and admit end of finger when scrotum is invaginated. Diagnosis: Chronic appendicitis, double hernia.

*Operative findings.*—No sac. Muscle deficiency.

*Operative procedure.*—Appendix removed through muscle-splitting incision. Operation on both sides completed as described.

*Result.*—April, 1938: Doctor reports no evidence of hernia. Since above note, patient returned to city for examination. No evidence of hernia found.

**Case 6.**—No. 40-89. March 27, 1935: Male, age 61, farmer and laborer.

*Duration.*—Five months.

*Physical examination.*—Right inguinal hernia; easily reducible; increasing in size. Has never extended into scrotum. No pain. No obstruction.

*Operative findings.*—Deficiency of muscle and conjoined tendon. Large defect. Mouth of sac extended nearly entire length of inguinal canal.

*Operative procedure.*—Isolation of sac, ligation, excision. Operation completed as described.

*Result.*—April, 1938: Doctor reports no evidence of hernia.

**Case 7.**—No. 40-97. April 2, 1935: Male, age 44, traveling agent.

*Duration.*—One and one-half years.

*Physical examination.*—Undescended testicle on right side. On standing has a large hernia which does not extend to scrotum. Reduces easily. Marked defect in external ring. On invagination of scrotum, pubic bone is easily palpated and finger passes into abdominal cavity.

*Operative findings.*—Large defect at lower margin of internal flat muscle, no conjoined tendon. Testicle in separate sac. Chronic appendicitis, adherent to neck of sac.

*Operative procedure.*—Appendix removed through muscle-splitting incision; sac isolated, removed, neck sutured over with fascia transversalis. Operation completed as described.

*Result.*—April, 1938: Doctor reports no evidence of hernia.

**Case 8.**—No. 40-138. May 9, 1935: Male, age 33, laborer.

*Duration.*—Fourteen or 15 years.

*Physical examination.*—Left inguinal hernia well down in scrotum. Difficult to reduce. Pubic bone easily palpated. External ring small.

*Operative findings.*—Lower segment of internal flat-muscles deficient and conjoined tendon consists of only a few fibers. Sac led up to internal ring.

*Operative procedure.*—Operation completed as described.

*Result.*—April, 1938: Doctor reports no evidence of hernia. Patient developed hernia on opposite side.

**Case 9.**—No. 40-158. May 21, 1935: Female, age 59, secretary.

*Duration.*—Twenty years.

*Physical examination.*—Truss discarded four years ago because ineffective. Hernia easily reduced. Pubic bone easily palpated.

*Operative findings.*—Conjoined tendon lacking. Hernia is fatty type with small sac originating at abdominal ring.

*Operative procedure.*—Sac isolated, ligated and removed. Operation completed as described.

*Result.*—April 22, 1938: Examined by myself; no evidence of hernia.

**Case 10.**—No. 40–274. October 19, 1935: Male, age 59, superintendent of coal mine

*Duration.*—One year.

*Physical examination.*—Left inguinal hernia. Does not enter scrotum. Large subcutaneous ring. Pubic bone easily palpated. Protrusion appears on standing. Disappears on lying down.

*Operative findings.*—Hernia came through immediately under subcutaneous ring. Only slight contact with cord. Entire absence of conjoined tendon.

*Operative procedure.*—Operation completed as described.

*Result.*—April, 1938: Examined by myself; no evidence of hernia.

**Case 11.**—No. 40–325. December 30, 1935: Male, age 34, truck driver; lifts heavy boxes containing metal.

*Duration.*—Many years.

*Physical examination.*—Right inguinal hernia. Massive incarceration extending one-third distance down thigh.

*Operative findings.*—Two segments of intestine, one obstructed. Reduced through right rectus incision. Conjoined tendon poorly developed. Sac isolated and neck sutured.

*Operative procedure.*—Operation completed as described.

*Result.*—April 21, 1938: Examined by myself; no evidence of hernia.

**Case 12.**—No. 41–26. January 25, 1936: Male, age 60, farmer and canner.

*Duration.*—Seven years.

*Physical examination.*—Left inguinal hernia. Massive incarceration. Syphilitic, paralyzed on right side, speech affected.

*Operative findings.*—Incarcerated, sliding hernia containing sigmoid with hypertrophied appendices epiploicae and mesosigmoid. Cord pushed down in front of sac. Enormous aperture one and one-half inches in diameter.

*Operative procedure.*—Sac incised below mesosigmoid, sutured and returned. Operation completed as described.

*Subsequent Course.*—Two weeks following operation developed a thrombosis of common iliac arteries and veins affecting both legs. Death from gangrene 57 days post operative. Autopsy demonstrated thrombi. Site of hernia excised for study: Hernia canal found obliterated. Pathologic examination of the fascial suture demonstrated that the histology was unaltered.

**Case 13.**—No. 41–61. February 29, 1936: Male, age 56, machinist.

*Duration.*—Some years.

*Physical examination.*—Right inguinal hernia. Indirect sac. On invagination of scrotum, finger passes directly over pubes into the abdominal cavity.

*Operative findings.*—Well-defined oblique sac. Conjoined tendon practically absent.

*Operative procedure.*—Sac isolated, ligated and excised. Operation completed as described.

*Result.*—April, 1938: Examined by myself; no evidence of hernia.

**Case 14.**—No. 41–77. March 26, 1936: Male, age 37, farmer and rural mail carrier.

*Duration.*—Eight years.

*Physical examination.*—Left inguinal hernia, incomplete. Hernia comes down in canal on coughing but is not complete. On invagination of scrotum, finger passes over pubic bone to its posterior surface.

*Operative findings.*—Sac of hernia had a very broad base alongside the cord, showing deficiency of posterior wall of canal. Conjoined tendon very poorly developed.

*Operative procedure.*—Sac isolated, excised and closed by suture. Operation completed as described.

*Result.*—Doctor reports no evidence of hernia.

**Case 15.**—No. 41–120. May 30, 1936: Male, age 20, laborer, works on roads.

*Duration.*—Four weeks.

*Physical examination.*—Right inguinal hernia. History of knot size of hen's egg, appearing while at work. On invagination of scrotum, finger passes over the pubic bone.

*Operative findings.*—No sac found. Conjoined tendon and lower segment of internal flat muscles absent.

*Operative procedure.*—Operation completed as described.

*Result.*—April, 1938: Doctor resports no evidence of hernia.

**Case 16.**—No. 41–131. June 3, 1936: Male, age 32, laborer.

*Duration.*—Six years.

*Physical examination.*—Right inguinal hernia. Truss ineffective. On invagination of scrotum, finger passes directly over the pubic bone.

*Operative findings.*—Congenital sac extending along cord. Central portion obliterated; below the sac is continuous with the tunica vaginalis. Conjoined tendon entirely absent, muscle fibers passing over directly to rectus sheath; external ring large.

*Operative procedure.*—Sac isolated, ligated and excised. Lower portion of sac isolated and constructed into a tunica. Operation completed as described.

*Result.*—April, 1938: Doctor reports no evidence of hernia.

**Case 17.**—No. 41–171. July 25, 1936: Male, age six.

*Duration.*—From birth.

*Physical examination.*—Right inguinal hernia extending into scrotum. Easily reduced. On invaginating scrotum, finger passes directly over pubic bone and into abdominal cavity.

*Operative findings.*—Definite sac extending to testicle. No abdominal ring. Neck of sac included whole of posterior wall of canal. Lower segment of internal flat muscles and conjoined tendon absent.

*Operative procedure.*—Sac removed. Operation completed as described.

*Result.*—April, 1938: Doctor reports no evidence of hernia.

**Case 18.**—No. 41–193. August 22, 1936: Male, age 65, dairyman.

*Duration.*—Four years. Four months ago hernia began to slip under truss.

*Physical examination.*—Right inguinal hernia. On invaginating scrotum, finger passes over pubic bone directly in for some distance.

*Operative findings.*—Cord had a sac running along it but contained no intestine. Lower segment of internal flat muscles and conjoined tendon lacking, producing a large defect behind external ring.

*Operative procedure.*—Sac isolated, ligated and excised. Operation completed as described.

*Result.*—April, 1938: Examined by myself; no evidence of hernia.

**Case 19.**—No. 41–284. November 28, 1936: Male, age 46, laborer and farmer.

*Duration.*—Some years.

*Physical examination.*—Double hernia. Right inguinal hernia, direct. Left inguinal hernia. Femoral hernia on right side and spermatocele on left. On invaginating scrotum, on both sides finger passes over pubic bone directly into abdominal cavity.

*Operative findings.*—No sac found on right; muscle deficiency. On left side, no conjoined tendon; muscle deficiency. Left aperture 2x2½ cm.

*Operative procedure.*—On right side, rectus sheath and muscle sutured to Cooper's ligament, behind femoral hernia; protecting both femoral and direct hernia. Operation completed as described on left side only.

*Result.*—April, 1938: Doctor reports no evidence of hernia.

**Case 20.**—No. 42–77. April 14, 1937: Male, age 48, captain in fire department.

*Duration.*—Two years.

*Physical examination.*—Right inguinal hernia. Protrusion extends half way to scrotum when standing. Cannot wear truss. Reduces when he lies down. Hernia is large. On invagination of scrotum, finger passes over pubic bone into abdominal cavity.

*Operative findings.*—Definite sac extending several inches along the cord. Conjoined tendon present but very attenuated.

*Operative procedure.*—Sac isolated, ligated and removed. Operation completed as described.

*Result.*—April, 1938: Doctor reports no evidence of hernia.

**Case 21.**—No. 42–86. April 16, 1937: Male, age 26, ambulance attendant.

*Duration.*—One month.

*Physical examination.*—Left inguinal hernia. Sudden onset of pain in left inguinal region while carrying patient downstairs on a stretcher. Protrusion occurred later. Has protrusion when standing; reduces when lying down. On invaginating scrotum, finger passes inward directly over pubic bone. Also has varicocele and elongated scrotum.

*Operative findings.*—Small sac at midpoint of Poupart's ligament. No conjoined tendon and muscle fibers pass directly to rectus sheath at a point about 5 cm. above crest of pubes.

*Operative procedure.*—Sac isolated and removed. Operation completed as described. Resection of pampiniform plexus, amputation of scrotum.

*Result.*—April, 1938: Examined by myself; no evidence of hernia.

**Case 22.**—No. 42–96. April 22, 1937: Male, age 28, sheet metal works.

*Duration.*—Five years. Second recurrence three months ago. First recurrence one month after operation. Second recurrence one and one-half months after operation.

*Physical examination.*—Direct hernia, left.

*Operative findings.*—Dissection showed only aperature was just above pubic bone, where the opening easily admitted the finger.

*Operative procedure.*—Scar tissue cut out. Tissues dissected out and demonstrated. Aperture closed by suturing rectus muscle and sheath to pectin ossis pubic with strip fascia from fascia lata.

*Result.*—April 30, 1938: Examined by myself; no evidence of hernia.

**Case 23.**—No. 42–245. November 11, 1937: Male, age 61, minister.

*Duration.*—Recent occurrence.

*Physical examination.*—On standing, has protrusion at external ring on left side, which reduces when he lies down. On invagination of scrotum, finger passes in directly over pubic bone.

*Operative findings.*—Long slender sac between pampiniform plexus and cord down to external ring. Accompanying the sac was a globular pad of fat. Finger passes directly through external ring to inside of pelvis.

*Operative procedure.*—Sac isolated, ligated and excised. Operation completed as described.

*Result.*—April, 1938: Examined by myself; no evidence of hernia.

*Discussion.*—The series in which the technic described in this paper was applied consisted of 23 patients, among whom there were four who had double herniae, making a total of 27 operations. This number of operations, while not large, were all performed by myself in an attempt to find out why herniae recur. In each case the tissues were thoroughly isolated by dissection and their insertion exposed, and the facts revealed studied in the light of what had been written on the subject.

The key to the diagnosis of direct hernia was the maneuver of invaginating the scrotum and passing the finger over the plainly felt superior surface of the pubic bone into the cavity of the pelvis beyond the insertion of the rectus muscle. The defect thus demonstrated was more marked in some cases than in others but was, however, definite in every case. In some of the cases two fingers could be introduced and swept around beyond the insertion of the rectus muscle.

I was able to demonstrate, in every case, that where this maneuver indicated that the conjoined tendon was deficient, on dissection it was found

to be absent. In addition to that, it was found that the lower bundles of fibers of the internal flat muscles were also absent. The widest gap above the pubic bone was 5 cm. (two inches), but the defect was not confined to the region of the external ring but extended lateralward to the internal ring or beyond. This probably explains why there are recurrences following operations for indirect hernia. It is now generally conceded that recurrence in these cases takes place below the internal ring.

The lower segment of the internal flat muscles and the conjoined tendon, which is a continuation of them, produce the shutter action referred to by Sir Arthur Keith,[17] so that there is not only a defect but a lack of this very important shutter action. I was able to demonstrate this very clearly in a case of indirect hernia with a rather large external ring but with an intact conjoined tendon. When the little finger was introduced into the inguinal canal and the patient coughed, the finger was constricted very firmly by the muscle.

In each case, notwithstanding the fact that the hernia was a definite direct one, the cord was examined carefully for a sac. In 14 of the 27 herniae a sac was found. Some were rudimentary, others larger. An interesting condition was found in three cases where the mouth of the sac was very large, corresponding to the length of the posterior wall of the inguinal canal.

*Follow-Up.*—All of the cases have been reexamined, either by the patient's attending physician or by myself, during the month of April, 1938. In every case the findings were the same. On invagination of the scrotum, the external ring was found to be firmly closed, so that not even the tip of the finger could be introduced. There was no bulging or protrusion at any point while standing, coughing or straining.

*Congenital Condition.*—From the unifomity with which the deficiency in the lower segment of the internal flat muscles and the absence of the conjoined tendon were found, it may be reasonably inferred that the predisposing cause for direct inguinal hernia is the presence of a congenital condition. This was borne out in this series of cases by the occurrence in three cases, whose ages were six, 19 and 20, respectively, in which it was noted that the lower segment of the internal flat muscles and the conjoined tendon were deficient. In the 19 and 20 year old cases, there was no sac. In the six year old case, there was a peculiar sac, in that there was no internal abdominal ring and the mouth of the sac included the whole of the posterior wall of the inguinal canal.

## CONCLUSIONS

(1) Direct inguinal hernia is caused by a deficiency of the lower segment of the internal flat muscle and the conjoined tendon.

(2) This appears to be a congenital condition, but as a rule the hernia does not appear until later in life.

(3) The defect can be closed with certainty only by the use of fascial suture.

(4) This defect presents the shape of an irregular triangle in which the base is the pubic bone. In closing this defect, the rectus muscle and the sheath must be sutured first to the ligamentous tissue covering the superior surface of the pubic bone, after which the internal flat muscles are sutured to Poupart's ligament, thus completing the closure.

(5) The operations described by Bassini, Halsted and Ferguson visualize hernia as being a sac protruding through the inguinal canal, and the operations are directed toward removal of the sac and the construction of a new and stronger canal for the cord. Direct hernia has no relation to the canal and the cord except that it appears at the lower end. The problem of curing direct hernia by one of these standard operations suggested modifications that have not been successful.

(6) The examination for inguinal hernia should always include invagination of the scrotum and exploration of the external ring and the superior surface of the pubic bone. If the finger passes directly over the pubic bone into the pelvic cavity, there is a defect which must be closed as described, irrespective of what else may be found necessary to be done for other conditions.

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## DISCUSSION OF THE PAPERS OF DOCTORS ROBINS AND WILLIAMS

DR. A. O. SINGLETON (Galveston, Texas): I wish to congratulate both essayists upon this excellent exposition of the repair of defects in the inguinal region. I would like to direct my remarks to Doctor Robins' paper. Doctor Robins has been thinking about these herniae for a long time, and has written upon the treatment of them previously. We know that where we have marked strain on the abdominal wall it is usually well supplied with fascia. As has been as well demonstrated, the great difficulty in repairing these defects is in the absence of fascia. About 1917, I read a paper reporting eight cases of transplanting some fascia in cases of inguinal hernia, and have practiced it on very difficult herniae, both direct and indirect, at different times. A few years ago the literature became filled with reports of fascial sutures, and I started employing these in place of fascial patches. Doctor Robins' suggestion of using this fascia has recalled some unpleasant experiences. I worked with these, trying to make and tie them, but it meant hours of work and not always satisfactory results, so I finally returned to the employment of the patches of fascia.

I looked up all the cases we have had and found there had been 54 transplants of fascial patches, in very difficult inguinal herniae, direct and indirect, selecting the difficult ones, as a rule those that had recurred. From the very first we practiced exposing the inguinal region, in preparation for repair, as one would by any other method. Under local anesthesia, a patch of fascia was removed from the thigh, after measuring the approximate size of the opening which was to be filled. A notch was cut in one end, and the cord placed through near the notch. It was then sutured along the inguinal ligament with fine chromic catgut silk and sutured carefully to the pubic bone. The fascia was then picked up with mattress stitches through the internal oblique muscle and these sutures were not tied at this time. The hernia was then repaired in the usual way, the muscle being brought down under the cord, after which the mattress stitches were tied. Following this the external oblique was brought together as one would do ordinarily. In other words, this transplant was placed behind the muscles, and then a Bassini hernia operation was performed over it. Out of 54 cases, selected from probably 1,200 herniae, extending over a period of some years, we have not been able to find one that has had a recurrence. We have had the

wound infected and drained for several days but no grafts have been lost. This method has, therefore, been found to be efficient for the cure of these defects in the inguinal region, and has not been followed by a recurrence in a single case.

DR. WILLARD BARTLETT (St. Louis, Mo.): I feel quite qualified to discuss this subject, I think, because of the fact that I have had more grief than anybody else with herniae. My outstanding result was on an old hospital orderly upon whom I operated for a bilateral recurrence after ten operations upon both sides for direct herniae. I am too modest to tell you what my result was, but he does not come in on the cases referred to above.

In this class of case, we adopted the following procedure for want of something better. We incised the skin, exposed the herniated mass, picked up the cord, pushed the herniated mass into the ring, and wove in fascia sutures like a chair bottom. We picked up the ring wherever we could get it, crisscrossed the sutures, and when completed, the individual could not push out his hernia at all. I do not remember a recurrence, though a number of cases are of long duration.

DR. CHARLES R. ROBINS (closing): I should like to say a word about the reference made to me in Doctor Williams' paper. One could not help but be pleased to realize that an expedient I had practiced some years ago should lead to the development of a standard operation for the cure of indirect hernia. The case in which I first performed the operation for the relief of hernia through an abdominal incision was to relieve a strangulated femoral hernia, which had been incarcerated for some time, and there was a possibility that it might be necessary to resect the intestine.

I had previously reduced a strangulated femoral hernia by incising Gimbernat's ligament, a procedure which had long been advocated. However, in this case, although the reduction resulted in the recovery of the patient from the strangulation, she subsequently developed a very large femoral hernia that could not be repaired.

I found that by approaching the hernia from above, through an abdominal incision, the reduction was simplified, and that it afforded ample space in which to resect the intestine, if necessary. I have approached the hernia from the abdomen in strangulation in a number of cases but have not adopted the procedure for the cure of simple hernia as a routine procedure. Doctor LaRoque developed the operation as a curative procedure, and Doctor Williams has still further developed it. One also sees very favorable references to the procedure in the literature.

Referring to Doctor Singleton's remarks, I have used the patch fascial transplant in a few cases with satisfactory results. The fascial suture is, however, a more convenient and more easily applied method of using the fascia. My first observation of the use of the fascial suture was at the Mayo Clinic some few years ago. Doctor Masson had devised a stripper, which I have found a most satisfactory instrument. However, I have not found it necessary to secure fascia from the thigh except when the pillars of the rings could not be used. The objection to getting fascia from the thigh, either patch or a suture, is that it makes two operations, and consequently prolongs the procedure.

The operation that I have shown is a simple one and can be done as quickly as when the prepared sutures are used. The only additional time required is to prepare the fascial strips and to attach them to the needle. This, however, is a matter of only a few minutes.

# MADELUNG'S DEFORMITY

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MADELUNG'S deformity may be described as an idiopathic, progressive curvature of the radius due to a dyschondroplasia of the inferior radial epiphysis, resulting in a deformity of the wrist, giving it the appearance of an anterior (or, more rarely, a posterior) subluxation of the hand. Several earlier authors, including Madelung himself, have accredited Dupuytren with the first reference to this condition, while others have given it the double patronym of "Dupuytren–Madelung," but Stetten,[221] who has carefully examined Dupuytren's right to this distinction, finds that it cannot be substantiated. Dupuytren's report,[73] in 1834, is based on a quotation taken, by his own admission, from Bégin,[11] who, in 1825, noted among adult male workers, not the typical spontaneous deformity, but a true forward dislocation of the wrist as a result of occupation. Other similar reports of a vague[213] or secondary static deformity[130] appeared soon after.

Probably the first description of the true deformity was that by Malgaigne,[138] in 1855, while in 1875 Jean [115] reported the first definite anatomic dissection. But the credit for first presenting a clear picture of it as a distinct clinical entity reverts to Madelung,[135] who, in 1878, before the Seventh Congress of German Surgeons, described the condition as a disturbance of growth in the joints, analogous to pes valgus, genu varum and scoliosis, and regarded it as a subluxation of the wrist joint. It remained for Duplay,[71] in 1885, to point out that the deformity was a result of volar bowing of the distal end of the radius.

Practically all that has been written on this subject has appeared in the French, German and Italian literature, and it was this fact, together with the paucity and meagerness of references to the subject in the various textbooks, that prompted Stetten,[221] in 1909, to describe, in American literature, a case of the deformity which he had previously reported[220] abroad, and at the same time presented a complete review of the literature relating to it. Although he definitely mentions several previously reported cases in America, he has been misrepresented by subsequent authors as having reported the first case from this country. At the present time one finds the reports of cases of this condition in a deplorable state. They are inadequate, often vague and unconvincing, while references are meager and all too frequently incorrect. Instead of being content with a case report, a quasi review of the

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literature has been presented which merely serves to perpetuate the inaccuracies of previous writers.

Pooley,[176] in 1880, evidently totally unaware of Madelung's report, described the first authentic case in American literature. Kieffer,[120] in 1902, briefly described another; Peckham,[165] in 1907, poorly presented a questionable case in a girl, age 14. Brinsmade,[28] in 1909, reported the fourth case; Stetten[221] wrote his incomparable article in 1909; a few months later Peckham and Hammond,[166] in a presentation of interesting cases from their clinic, reported two cases (Nos. 3 and 4) as examples of Madelung's deformity. In one of these no evidence or description warranting a definite diagnosis of the genuine deformity is given, although the other case (No. 3) is an excellent example of the rare reverse type of the deformity. Stokes[223] added two more American cases in 1910, while in 1911, Jones[116] briefly described another case, and Taylor[225] reported two cases in 1912. In 1914, Adler[1] briefly described a case which he called Madelung's deformity, while in 1915, Parkes[164] reported a case upon which he had operated with excellent results. In 1916, Earl[74] gave a good presentation of an early case of the deformity.

Despite the availability of these reports in American literature, or at least their mention in Stetten's article, Brown,[31] in 1924, reported his case as "the fourth case reported in America," while in the same year Levyn,[132] realizing Brown's mistake, as he thought, wrote a "report of two cases constituting the fifth and sixth American cases," when in truth they were really the sixteenth and seventeenth. Also in 1924, Moore[154] reported two cases upon which he had operated, while in 1936, Claiborne[43] reported a case. Emboldened by a thorough, methodical and exact survey of the related American literature, we feel we are correct in reporting the case herein cited as the twenty-first from this country. Two reports[162, 238] have not been included in this series since they obviously do not fall into the category of a true Madelung's deformity, presenting, as they both do, lesions of the ulna and not of the radius.

Comprehensive reviews on this subject have appeared from time to time. In 1903, Abadie[1] compiled a bibliography reporting 41 known cases of the deformity which he had succeeded in collecting. Gasne[95] reviewed the subject in 1906, but did not add any new cases. In 1907, Estor[77] published a total of 85 cases. Stetten[221] collected, and gave brief summaries of 62 cases up to 1908. In 1908, Siegrist[212] tabulated 58 cases, and Franke,[87] only 56. In 1909, Marsan[140] listed 90 cases; in 1911, Ramos[185] listed 69 cases, while in 1913, Melchior,[152] after reviewing the subject, accepted only 75 cases, while in 1933, Salisachs[203] reported a total of 133 known cases

The discrepancy in these figures is obvious, and is due to the considerable difference of individual opinion as to what constitutes a true instance of this deformity and on what evidence it is to be accepted. Although only five cases reported by Madelung are specific enough for acceptance, he claims to have seen 12, and he is sometimes credited with that number. In the discussion of Madelung's paper,[135] Czerny claimed to have seen two cases,

Hirschberg two cases, and similar cases were claimed to have been noted by Langenbeck, but Abadie[1] discards their claims, being unable to find published reports of their observations.

In the discussion of Gangolphe's paper,[92] Bérard[15] described a case of his own, which Stetten is inclined to accept as genuine, but which we have discarded as being too indefinite for acceptance. After describing a case occurring in mother and daughter, Guépin[104] reports a similar deformity in 14 other members of the family, while Féré[83] claims to have seen 25 cases in male epileptics. We are rather reluctant to embody these figures into our own statistics. We have found, as Stetten also remarks, considerable difficulty in knowing just which cases one should incorporate. In our own tabulation, we have tried to follow Stetten's criteria in rigidly rejecting all cases where:

(1) There is a reasonable doubt as to the actual existence of a typical Madelung's deformity, and a probability that the condition was some other pathologic lesion.

(2) The deformity was of such a slight degree that it could scarcely be considered abnormal.

(3) The description was too vague or the reference too indefinite to justify inclusion.

Also tending to upset an accurate statistical compilation is the occasional report of the same case by two men, as it occurs in the articles by Weber[235] and Busch,[35] and Müller[155] and Franke.[87] Stetten himself has reported the same case in two journals,[220, 221] while it has been a common practice for several of the authors to rearrange their primary case report in subsequent articles.

In view of the foregoing, we have attempted to eliminate these inaccuracies from the literature, and have compiled a chart tabulating 171 cases which we have succeeded in collecting.

We cannot agree with Claiborne[43] in assuming that with the increasing number of examinations which are resulting from the Workmen's Compensation Act, and with the wider use of roentgenologic examinations in traumatic surgery, there will be more observations and reports of this condition, since the type of patient presenting a true Madelung's deformity does not come from either of these two groups. We do believe, however, that the condition is not so rare as the literature would seem to indicate, since it is not frequently recognized, especially without the aid of the roentgenologist.

**Case Report.**—Hosp. No. 69356: M. D., female, age 18, was admitted to the Cumberland Hospital, January 31, 1938, to the service of Dr. G. B. Reitz, complaining of pain in, and deformity of, both wrists during the past two years. She was born April 1, 1920, in the United States, and no history was obtainable of any birth injury having occurred. She had had measles, whooping cough and diphtheria. At age 9, she was struck by an automobile, but there was no history or evidence of any local injury to the wrists. She had not engaged in any particular occupation which might account for her deformity. Her mother, age 47, three sisters and two brothers are all living and well. Her father died of pneumonia at age 47. There were no deformities or any

indication of syphilis or rickets in either parent or in any of her five brothers and sisters.

*Physical Examination* revealed a well developed, well nourished girl, 160 cm. tall, weighing 50 kg. Temperature, 99.2° F.; pulse, 84; respirations, 16; blood pressure, 130/65. The visible mucous membranes are of good color and the teeth are in good condition.

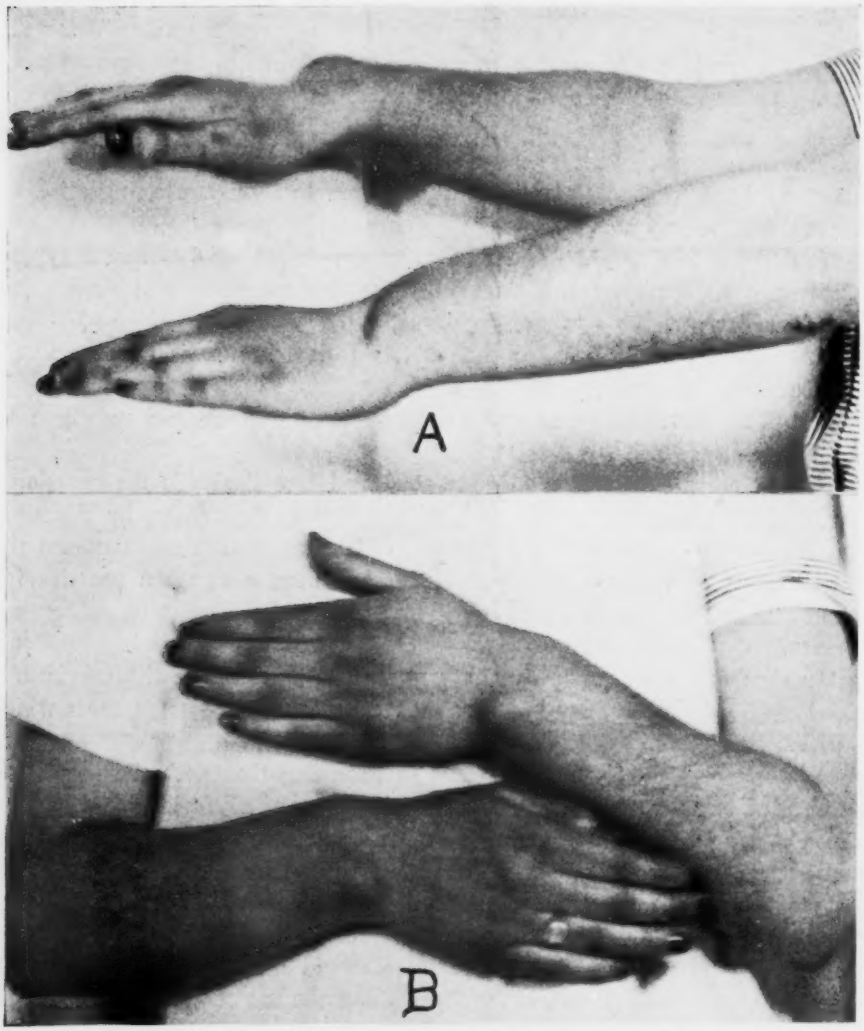


FIG. 1.—(A) Lateral view showing the characteristic bayonet-shaped deformity and the shortening in the length of both forearms.
(B) Anteroposterior view showing the prominence of the head of the ulna at the back of the wrist, and the ulnar deviation of the hand.

The organs of special sense, and the thoracic and abdominal viscera are all apparently normal, and there are no sensory, trophic or vasomotor disturbances. There are no frontal or parietal bosses, the thorax is well formed and there is no scoliosis, rachitic rosary or Harrison's groove. The hips and knees are normal and the tibiae are perfectly straight. There is no broadening of the epiphyses.

One is immediately struck by a curious bilateral deformity at both wrists (Fig. 1 A

TABULATION OF ANALYSES OF RELEVANT DATA OF 171 CASES OF TRUE MADELUNG'S DEFORMITY

| No. | Author and Year | Age and Sex | Occupation | Age at Onset | Casual Relationship | Heredity | Unilateral or Bilateral | Wrist Joint: Symptoms | Wrist Joint: Function | Examination | Other Abnormalities | Treatment | Course or Result |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 1. | Malgaigne 1855 | 36 M. | Mason | 10–12 | Fracture middle of radius 3 yrs. before onset | — | Unilateral | Pain at onset for 18 mos. | Good, except for extension and pronation | Radius: Bowed at lower extremity | Exostosis of radius, just above wrist | Unsuccessful attempt at reduction | Progressed 18 mos. |
| 2. | Weber 1859 | 16 F. | — | 12–15 | No history of trauma | — | Right | Pain at onset and on motion | Extension limited | Radius and ulna: Project at back of wrist. Carpus: Apparently displaced anteriorly | — | Reduction under narcosis but recurrence. Reduced and maintained with plaster encasement and apparatus | Gradual progress (was later operated upon by Busch—see below) |
| 2. | Busch 1864 | 21 F. | — | 12–15 | No history of trauma | — | Bilateral | Pain on working | Limitation of motion | Apparent anterior displacement of the hand | — | Tenotomy of the flexor carpi ulnaris and palmaris longus tendons | Result poor; progression |
| 3. | Jean 1875 | 73 F. | — | Childhood | Congenital? | — | Bilateral, more marked on right | — | Extension limited | Radius: Lower extremity bowed forward. Ulna: Dislocated backward. Carpus: Displaced forward | Hyperostosis of the head of the ulna | *Autopsy Dissection* | |
| 4. | Madelung 1878 | 20 F. | — | Childhood | No history of trauma | — | Left | — | — | Carpus and hand: Dislocated forward on wrist. Radius: Bowed forward at epiphysis. | — | *Autopsy Dissection* | |
| 5. | Madelung 1878 | 18 F. | Peasant | 15 | — | — | Bilateral | Pain on extension | Interference with work | Apparent forward dislocation of hand and carpus | — | Avoidance of extension and hard work. Leather wrist bracelet | Pain relieved |
| 6. | Madelung 1878 | 16 F. | Wealthy family | — | — | — | Right | Pain on extension | — | — | — | Leather wrist bracelet | Pain relieved |
| 7. | Madelung 1878 | 14 M. | Tanner | — | Lifted heavy weights | — | Bilateral | Pain during development | Extension limited | Prominence of the head of the ulna at the back of the wrist | — | Avoided heavy work. Leather wrist bracelet | Pain relieved |
| 8. | Madelung 1878 | 15 F. | — | Childhood | — | — | Left | Pain on extension | Extension limited | Marked subluxation forward of carpus and hand | — | Leather wrist bracelet | Pain relieved |
| 9. | Pooley 1880 | 33 F. | Dairymaid | 8 | No history of trauma or local disease | — | Bilateral | Pain on milking cows and on hard work | Extension limited; flexion increased | Ulna: Styloid prominent. Radius and ulna: Apparently shortened. Carpus: Dislocated forward | Hyperostosis of the ulnar head | — | — |
| 10. | Felix (Tripier) 1884 | — F. | Laundress | 14 | — | — | Bilateral, more marked on left | Severe pain at onset | Motion disturbed; supination impossible | Ulna: Dislocated backward. Carpus and hand: Subluxated forward | — | Orthopedic appliance; electricity | Recurrance of deformity |
| 11. | Duplay 1885 | — F. | — | 9 | Strained wrist 1–2 yrs. before | — | Left | — | No loss of motion | Radius: Lower end bent forward. Ulna: Lower end dislocated backward. Carpus and hand: Apparent subluxation forward | Scoliosis. Hyperostosis of the ulnar head | Failure of orthopedic appliance. Linear osteotomy 3 cm. above joint | Result: Good |
| 12. | von Bergmann 1888 | — F. | — | Childhood | No history of trauma | — | Bilateral | Severe pains at onset | Extension limited | Apparent forward subluxation of carpus and hand on wrist | Bilateral shortening of the fourth metacarpals | — | Gradual progression |
| 13. | Duplay 1891 | 14 F. | — | 7 | — | — | Bilateral, more marked on right | Easily fatigued. Vague pains at onset | Slight disturbance of motion | Radius: Lower end bent forward. Ulna: Slightly bent forward. Carpus and hand: Displaced anteriorly | Hyperostosis of the ulnar head | — | Course: 4 to 6 mos. |
| 14. | Bode 1891 | — F. | — | 13 | — | — | Left | — | — | Ulna displaced forward (?) | Hemiatrophy of the right side | Deformity reducible on pressure | Recurs |
| 15. | Bennett 1891 | 24 F. | Cook | 23 | No trauma. No rheumatism | — | Bilateral, especially the right | Increasing weakness of wrists; must stop work | — | Ulna: Dislocated backward from radius and carpus. Carpus and hand: Displaced anteriorly | — | Deformity on right side reducible by pressure | — |
| 16. | Guepin 1892 | — F. | — | Childhood | Slight trauma to left wrist as an infant | Mother, father, brothers, sisters, uncles and aunts, nephews and nieces (14 members of the family) have similar deformities | Bilateral, especially the right | — | — | Right radius: Completely dislocated forward from the ulna. Displacement of the carpus from the ulna complete, and from the radius nearly so. Articular ends of the radius and ulna project under the skin. Left radius: Displacement only partial representing an early phase of the deformity on the right | — | Deformity of both sides can be reduced by pressure, under anesthesia | Spontaneous, immediate recurrence of the deformity |
| 17. | Guepin 1892 | — F. | — | — | — | Mother of above | Bilateral | — | — | Ulna: Dislocated backward from radius and carpus. Carpus and hand: Dislocated from ulna, and subluxated from the radius | — | — | — |
| 18. | Redard | — P. | Washwoman | 14 | No rickets. History of fall on hand | — | Bilateral | Active pain especially on wringing wash | Motion limited, especially extension and supination | Radius: Marked forward bowing. Ulna: Slightly bowed forward. Carpus and hand: In position of subluxation | No stigmata of rickets | Rest; galvanism | — |
| 19. | Malfuson 1894 | — P. | Seamstress | 13 | History of having wrenched hands | Mother has same deformity | Bilateral, more marked on right | Intermittent pain | Extension limited and supination painful on right side only | Radius: Slight bowing of the lower quarter of shaft. Carpus and hand: Subluxated forward; and hand displaced toward the ulna | Mild scoliosis | Massage; sulphur baths; iodine | — |
| 20. | Malfuson 1894 | — F. | — | — | — | Mother of above | Bilateral | No pain | No disturbance of function | Radius: Bowed forward. Carpus and hand: Subluxated at wrist | — | — | — |
| 21. | Jagot 1897 | 21 F. | Milkmaid | 13–14 | Hard work | Father, uncle and grandfather have large wrists | Bilateral | No pain | Extension and lateral motion limited | Radius: Bowed forward. Ulna: Dislocated backward. Carpus and hand: Displaced forward. Roentgenogram | Hypertrophy of lower third of the ulna | — | — |
| 22. | Nélaton 1897 | — | — | — | — | — | Unilateral | *Dissected Specimen* | | Radius: Bowed forward. Carpus and hand: Displaced anteriorly | — | *Dissected Specimen* | |
| 23. | Guéry 1898 | — M. | Carpet-weaver | 13 | History of strain to wrist | — | Left | Marked pain in wrist | Flexion and abduction increased; extension and adduction diminished | Radius: Articular surface deviated anteriorly. Ulna: Dislocated backward. Carpus and hand: Displaced forward and to the ulnar side, but in normal relation to the radius. Roentgenogram | Hyperostosis of the head of the ulna | Plaster encasement 2 wks., with hand in extension and half pronation. Leather bracelet | Recurrence after reduction, but gradual improvement |
| 24. | Delbet 1899 | 16 F. | — | 12 | Denies any history of trauma | — | Bilateral | Pain | Disturbance in ability to write | Radius: Bowed forward at lower end. Ulna: Dislocated backward. Carpus and hand: Displaced anteriorly and toward ulna | — | Linear osteotomy of the lower end of the radius. Plaster encasement | Course: 4 yrs. Tendency to recurrence |
| 25. | Cabeca 1899 | — F. | Pianist | 14 | No history of trauma | — | Bilateral, mostly right | Pain at the beginning | Normal motion | Radius: Bowed forward at lower end. Ulna: Dislocated backward, Carpus: Displaced forward. Roentgenogram | — | — | — |
| 26. | Gangolphe 1899 | — F. | Embroiderer | 14 | No history of trauma. No stigmata of rickets | Mother has same deformity | Bilateral, especially right | Severe pain | Extension limited on right side | Radius: Bowed forward and toward ulna at lower end. Ulna: Dislocated backward. Roentgenogram | Slight scoliosis. Lax ligaments of left knee | — | — |
| 27. | Gangolphe 1899 | — F. | Seamstress | 11 | No stigmata of rickets. No history of trauma | Mother of above | Bilateral, especially right | Easily fatigued; occasional pain | Extension, pronation and supination limited, especially on right | Radius: Bowed forward and toward ulna at lower end; right radius 3 cm. shorter than left. Ulna: Dislocated backward, right side shorter than left. Carpus and hand. Displaced forward, and to ulnar side. Roentgenogram | — | — | — |
| 28. | Roget 1899 | — F. | — | 8–10 | No trauma. No rickets | Mother and grandfather have prominent ulnae | Bilateral, especially right | Fatigue on writing | — | Radius: Bowed forward and toward ulna at lower end. Ulna: Dislocated backward. Carpus and hand: Displaced forward and held in adduction | Cubitus valgus | — | Course: 6 yrs. |
| 29. | Dekeyser 1901 | — F. | — | 14 | No history of trauma. No hard work | Mother has similar deformity | Right | — | Flexion increased | Radius: Shortened and lower end bent forward. Ulna: Dislocated backward. Carpus and hand: Displaced forward from ulna, in normal relation to radius. Roentgenogram | — | Rest | — |
| 30. | Dekeyser 1901 | — F. | — | Childhood | — | Mother of above | Left | Fatigue, weakness, clumsiness | Flexion slightly diminished on left | Radius: Shortened and bowed forward. Ulna: Dislocated backward. Carpus and hand displaced forward | — | — | Gradual progression |
| 31. | Kirmisson 1902 | 10½ F. | — | 10 | No rickets or trauma | — | Bilateral, mostly right | Fatigue; pain on flexion | Flexion diminished; abduction impossible; supination limited; extension and adduction increased | Radius: Lower end bowed back and toward ulna, shortened 3 cm. Ulna: Displaced forward, also shortened. Carpus and hand. Displaced backward, in normal relation to radius. Roentgenogram | Right dorsal scoliosis with curvature of right ribs and right scapula | Linear osteotomy of radius with resection of overriding piece of ulna. Leather bracelet | Good result |
| 32. | Gevaert 1902 | — M. | Weaver | 14 | Work entails continuous traction | — | Bilateral | Fatigue, weakness and clumsiness | Flexion slightly diminished | Radius: Bent forward at lower end. Ulna: Displaced backward. Carpus and hand: Displaced forward, in normal relation to radius. Roentgenogram | Dorsal scoliosis. Compensatory lumbar scoliosis. Genu valgum | Massage; electricity; passive motion | Course: 2 yrs. Result satisfactory |
| 33. | Ardouin 1902 | — F. | Waitress | Childhood | No history of trauma | Brother has large wrists | Bilateral, more marked on left | Slight weakness | — | Radius: Bowed forward, shortened. Ulna: Dislocated backward, shortened. Carpus and hand: Displaced forward, in normal relation to radius. Roentgenogram | Slight scoliosis | — | — |
| 34. | Abadie 1903 | 28 M. | Cook | 13 | Fall on hand at age 9, with possible fracture | — | Left | Weakness | All movements limited, especially pronation and supination | Radius: Lower end bowed forward. Ulna: Bowed in to radius and dislocated backward. Carpus and hand: Displaced forward, in normal relation to radius. Roentgenogram | Exostosis on left radius | — | Course: Several years |
| 35. | Cnopf 1903 | 15 F. | — | 12 | No trauma or hard work | — | Right | No pain | Extension slightly limited | Radius: Shortened, dorsal concave bowing in upper third, moderate volar concavity in lower two-thirds. Sharp bend forward at lower end, deviation of joint surface toward ulna. Carpus and hand: Displaced forward. Roentgenogram | — | — | Course: 2½ yrs. |
| 36. | Kieffer 1903 | 30 M. | Soldier | Infancy | Strenuous athletics | — | Bilateral, more marked on left | No pain | Not impaired | Ulna: Marked prominence of the styloid process at the back of the wrists, overriding the carpus dorsally, and subluxated from the radius and carpus. Roentgenogram | — | — | — |
| 37. | Bennecke 1904 | — F. | Printer | 15 | History of carrying heavy weights | | Bilateral, especially the right | Pain at onset, and while at work | Extension limited; pronation and supination diminished | Radius: Bowed forward. Ulna: Dislocated backward, both shortened. Carpus and hand: Displaced anteriorly. First row of carpal bones wedge-shaped. Roentgenogram | Ulnar hyperostosis; slight scoliosis; cubitus valgus (right) | — | — |
| 38. | Poulsen 1904 | — F. | — | 12 | — | — | Left | No pain | Extension and supination limited | Radius: Bent forward sharply turning joint surface in, shortened. Ulna: Displaced backward, shortened. Carpus and hand: Displaced forward and to radius. Roentgenogram | — | Oblique linear osteotomy. Bandaged in extension | Course: 4 yrs. Tendency to recur |
| 39. | Poulsen 1904 | — F. | Servant | 12 | — | — | Bilateral, more marked on left | No pain | Slight limitation of supination | Radius: Shaft bent forward, especially at lower end, and also bowed toward ulna, with deviation of the joint surface. Ulna: Displaced backward. Carpus and hand: placed forward in normal relation to radius. Roentgenogram | — | Oblique linear osteotomy. Bandage | Course: 1 yr. Improvement |
| 40. | Albertin and Leclerc 1905 | — P. | Corsetmaker | Childhood | Both arms pulled violently at age 5 | — | Bilateral | Pain and cramps on overwork | Flexion and abduction slightly exaggerated | Radius: Curved forward. Ulna: Dislocated backward on radius and carpus. Hand: Apparently dislocated forward. "Pyramidalization" of first row of carpus | — | Ulnar dislocation can be reduced by pressure | Recurs |
| 41. | Schulze 1905 | — F. | — | 9 | Hand twisted at play | — | Right | No pain. Easily fatigued, especially playing piano | Extension limited; adduction slightly; abduction considerably | Radius: Lower half bowed forward. Ulna: Displaced backward and elongated. Hand: Displaced forward | Cubitus valgus. Congenital dislocation of both hips | Massage; electricity. Bandaged in extension | — |

| | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | | | and supination | ward. Carpus and hand: Displaced forward, in normal relation to radius. Roentgenogram | | | |
| 35. Cnopf 1903 | 15 F. | — | 12 | No trauma or hard work | — | Right | No pain | Extension slightly limited | Radius: Shortened, dorsal concave bowing in upper third, moderate volar concavity in lower two-thirds. Sharp bend forward at lower end, deviation of joint surface toward ulna. Carpus and hand: Displaced forward. Roentgenogram | — | — | Course: 2½ yrs. |
| 36. Kieffer 1903 | 30 M. | Soldier | Infancy | Strenuous athletics | — | Bilateral, more marked on left | No pain | Not impaired | Ulna: Marked prominence of the styloid process at the back of the wrists, overriding the carpus dorsally, and subluxated from the radius and carpus. Roentgenogram | — | — | — |
| 37. Bennecke 1904 | — F. | Printer | 15 | History of carrying heavy weights | | Bilateral, especially the right | Pain at onset, and while at work | Extension limited; pronation and supination diminished | Radius: Bowed forward. Ulna: Dislocated backward, both shortened. Carpus and hand: Displaced anteriorly. First row of carpal bones wedge-shaped. Roentgenogram | Ulnar hyperostosis; slight scoliosis; cubitus valgus (right) | — | — |
| 38. Poulsen 1904 | — F. | — | 12 | — | — | Left | No pain | Extension and supination limited | Radius: Bent forward sharply turning joint surface in, shortened. Ulna: Displaced backward, shortened. Carpus and hand: Displaced forward and to radius. Roentgenogram | — | Oblique linear osteotomy. Bandaged in extension | Course: 4 yrs. Tendency to recur |
| 39. Poulsen 1904 | — F. | Servant | 12 | — | — | Bilateral, more marked on left | No pain | Slight limitation of supination | Radius: Shaft bent forward, especially at lower end, and also bowed toward ulna, with deviation of the joint surface. Ulna: Displaced backward. Carpus and hand: placed forward in normal relation to radius. Roentgenogram | — | Oblique linear osteotomy. Bandage | Course: 1 yr. Improvement |
| 40. Albertin and Leclerc 1905 | — F. | Corsetmaker | Childhood | Both arms pulled violently at age 5 | — | Bilateral | Pain and cramps on overwork | Flexion and abduction slightly exaggerated | Radius: Curved forward. Ulna: Dislocated backward on radius and carpus. Hand: Apparently dislocated forward. "Pyramidalization" of first row of carpus | — | Ulnar dislocation can be reduced by pressure | Recurs |
| 41. Schulze 1905 | — F. | — | 9 | Hand twisted at play | — | Right | No pain. Easily fatigued, especially playing piano | Extension limited; adduction slightly; abduction considerably | Radius: Lower half bowed forward. Ulna: Displaced backward and elongated. Hand: Displaced forward | Cubitus valgus. Congenital dislocation of both hips | Massage; electricity. Bandaged in extension | — |
| 42. Volkmann 1905 | — F. | — | 16 | — | Younger sister has indication of same deformity | Bilateral | No pain | Flexion and supination limited | Radius: Lower third bowed forward and toward ulna, shortened. Ulna: Dislocated backward, shortened. Carpus: Displaced anteriorly, in relation to articular surface of the radius. Roentgenogram | — | None | Irreducible |
| 43. Volkmann 1905 | — F. | — | 14 | — | Mother and grandmother have same deformities | Bilateral, especially the right | No pain | No serious disturbance of motion | Radius: Bowed. Ulna: Dislocated backward. Carpus and hand: Displaced anteriorly. Roentgenogram | Cubitus valgus; pes planus | None | Ulna reducible on pressure but recurs |
| 44. Natvig 1905 | — M. | — | 14 | Weak as an infant | — | Bilateral more marked on right | Pain at onset | Extension and supination limited | Carpus: Displaced forward | — | — | — |
| 45. Solberg 1906 | 34 M. | Policeman | 16½ | — | Parents were cousins. Brother has similar deformity | Bilateral, especially the right | Pain at onset, and fatigue on doing heavy work | No limitation of motion | Ulna: End prominent. Carpus: Displaced forward | Slightly dwarfed; slight genu varum and flatfoot. left | — | — |
| 46. Solberg 1906 | 40 M. | — | 15 | — | Older brother of above | Right | No pain | — | Carpus: Displaced forward | Slightly dwarfed, and slight genu varum | — | — |
| 47. Piollet 1906 | 12 F. | Shopgirl | 8 | — | — | Bilateral | Pain in beginning | Extension limited | Radius: Lower third bowed forward. Ulna: Dislocated backward. Carpus and hand: Displaced forward. Roentgenogram | — | Orthopedic apparatus | Course: 4 yrs. No improvement |
| 48. Schade 1906 | 8 F. | — | — | No history of trauma | — | Bilateral, especially the right | No pain | Extension limited; abduction impossible; flexion increased | Radius: Lower third bowed forward and inward. Ulna: Dislocated backward on radius and carpus. Hand and wrist: Displaced forward, in normal relation to radius. Roentgenogram | Slight rosary and scoliosis | — | — |
| 49. Putti 1906 | 15 F. | Housework; washing | — | No history of trauma | — | Left | All movements painful | Extension limited | Radius: Lower third bent forward and inward. Ulna: Dislocated back and bent slightly forward. Carpus: Displaced forward, in normal relation to radius. Roentgenogram | Cubitus valgus | Oblique linear osteotomy of radius. Plaster encasement | Course: 1 yr. Good result |
| 50. Sauer 1906 | — F. | Servant | Childhood | No history of trauma | Several sisters and brothers said to have similarly deformed wrists | Bilateral | No pain | Extension limited 135°; abduction almost gone; flexion and adduction increased | Radius: Lower half bowed forward and toward ulna, especially at the epiphysis; shortened; interosseous crest prominent. Ulna: Displaced backward. Carpus and hand: Displaced forward with radius. Roentgenogram | Hyperostosis of the head of the ulna. Funnel chest | Avoidance of forced flexion. Exercise | Deformity irreducible |
| 51. Sauer 1906 | 18 F. | Servant | — | No history of trauma | — | Left | Easily tired, especially in wringing wash | Extension diminished; flexion increased | Radius: Normal (?) with broadening of the epiphyseal line. Ulna: Dislocated backward. Carpus and hand: Displaced forward along with the radius | Pes planus, and bilateral genu varum | — | Course: 1 yr. |
| 52. Sauer 1906 | 20 F. | Servant | — | — | — | Left | Pain when doing hard work | No disturbance of motion | Radius: Bowed forward at lower end, shortened. Ulna: Dislocated backward. Carpus and hand: Displaced forward in normal relation to radius. Roentgenogram | — | — | — |
| 53. Stieda 1907 | — F. | — | — | — | — | — | — | — | Radius: Bowed forward. Carpus and hand: Subluxated forward. Roentgenogram | — | — | — |
| 54. Lenormant 1907 | 13 F. | — | Early childhood | Rheumatism at 8 yrs. No trauma or rickets | — | Right | Pain and weakness | Extension limited; flexion increased | Radius: Shortened: lower fourth bowed forward, deviating the joint surface. Ulna: Dislocated backward. Hand and carpus: Displaced forward with the radius. Roentgenogram | Dorsal scoliosis. Depression of xyphoid cartilage | Osteotomy of the radius | Good result |
| 55. Pels-Leusden 1907 | — F. | Servant | 14 | Hard work | — | Bilateral, especially the right | Pain, especially in right wrist | Slight diminution of extension and abduction | Radius: Forward and medial bowing, with deviation of joint surfaces: right radius shorter than left. Ulna: Dislocated backward. Carpus and hand: Displaced forward. Roentgenogram | — | — | — |
| 56. Pels-Leusden 1907 | 20 F. | — | 12 | — | — | Bilateral | — | Motion limited | Radius: Bowed forward and toward ulna with deviation of the joint surface. Epiphyseal lines unfused (20 yrs.). Ulna: Displaced backward. Roentgenogram | Exostosis of radius proximal to epiphysis | — | — |
| 57. Estor 1907 | — F. | — | Birth | No history of trauma | Father has similar deformity | Bilateral, especially the right | — | No disturbance of motion | Radius: Bowed slightly inward and markedly forward. Ulna: Dislocated backward on radius and carpus, lower fourth bowed outward and backward. Carpus and hand: Displaced forward. Os magnum prominent. Roentgenogram | — | Ulna can be reduced by strong pressure | Recurrence |
| 58. Peckham 1907 | 17 F. | — | 14 | — | — | Bilateral | — | — | Radius: Bowed forward. Ulna: Dislocated backward. Carpus and hand: Displaced forward. Roentgenogram | — | Open reduction by dissecting out intervening fibrous tissue at wrist | Improved |
| 59. Siegrist 1908 | 18 F. | — | 11 | No trauma or heavy work. Plays piano. No rickets | Maternal grandmother, mother and two sisters have similar deformities | Bilateral, more marked on the left | Pain at onset of the deformity | Flexion, extension, adduction, abduction and supination diminished, especially on the left | Radius: Bowing of the shaft forward and inward, especially at the lower end. Ulna: Displaced backward. Carpus and hand: Displaced forward and toward ulna. First row of carpal bones wedge-shaped. Roentgenogram | Exostosis of left radius. Dorsal scoliosis. Pes valgus | Orthopedic appliance | No improvement |
| 60. Siegrist 1908 | 20 F. | Salesgirl | 14 | No history of trauma or heavy work | Sister of above and grandmother have similar deformities | Bilateral, more marked on left | Severe pain until height of deformity was reached | Extension limited and flexion increased | Radius: Moderate forward bowing of the entire shaft. Ulna: Dislocated backward. Carpus and hand: Displaced forward. Roentgenogram | Scoliosis and left pes planovalgus | — | — |
| 61. Siegrist 1908 | 44 F. | Washwoman | 10–12 | No history of trauma or heavy work | Mother of above two cases | Bilateral, especially the right | Severe pain at beginning, and on certain movements | All movements somewhat limited | Radius: Shaft shortened and bowed forward and inward at the lower end; deviation of the joint surface on two axes. Ulna: Displaced backward. Carpus and hand: Forward. Roentgenogram | — | — | — |
| 62. Franke (Müller) 1908 | 37 F. | — | Early childhood | History of rickets as a child | — | Bilateral | No pain | Extension and abduction limited | Radius: Bowed toward ulna at middle, and forward at lower end; shortened; has exostosis. Ulna: Displaced backward; shortened. Carpus and hand: Displaced forward. Angulation of the first row of carpal bones, second row extended. Roentgenogram | Dwarfism. Exostosis tibia and fibula. Rosary | *Necropsy Dissection* | |
| 63. Brinsmade 1908 | 15 F. | — | 12 | Arms twisted in childhood | — | Bilateral, more right | — | — | Radius: Lower end bowed forward and lower half bowed toward ulna. Ulna: Displaced backward. Carpus and hand: Forward. Roentgenogram | Slight scoliosis. Tibial bowing | — | Course: 1¾ yrs. |

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TABULATION OF ANALYSES OF RELEVANT DATA OF 171 CASES OF TRUE MADELUNG'S DEFORMITY (*Continued*)

| No. | Author and Year | Age and Sex | Occupation | Age at Onset | Casual Relationship | Heredity | Unilateral or Bilateral | Wrist Joint | | Examination | Other Abnormalities | Treatment | Course or Result |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | | | Symptoms | Function | | | | |
| 64. | Stetten 1908 | — F. | Schoolgirl | 11½ | No rickets. History of fall after onset of the deformity | Mother presents prominent ulnae | Bilateral, more marked on the left | Slight pain on the right side | Flexion diminished; abduction almost nil on left, restricted on right; adduction and extension increased; pronation and supination limited | Radius: Lower half bowed backward and toward ulna, shortened. Ulna: Dislocated forward with exaggeration of forward curvature; shortened. Carpus and hand. Displaced backward and toward ulna. Angulation of the first row of carpal bones. Roentgenogram | Exostosis of left radius | — | — |
| 65. | Denucé et Rabere 1908 | 11 M. | — | 10 | Spontaneous | — | Bilateral | No pain | Extension is limited. Flexion is exaggerated | Ulnae: Prominence of the heads at the back of the wrists. Radius: Anterior curvature of the distal ends. Hands and carpus: Anterior displacement. Roentgenogram | — | Radio-ulnar luxation, reducible on pressure | Recurrence |
| 66. | Marsan 1908 | 19 M. | Joiner and carriage-maker | 16 | Hard work as a boy. No signs of rickets | — | Bilateral, more marked on left | Pain very severe at onset, still present on forced motion. Weakness | Flexion good. Extension zero. Pronation and supination limited | Ulna: Swelling at back of wrist due to luxation of ulnar head from radius and carpus. Hand and carpus: Apparent anterior displacement. Radius: Shortened, bowed anteriorly. Roentgenogram | Tuberculous arthritis of left ankle | — | — |
| 67. | Peckam and Hammond 1909 | 13 M. | — | 11 | No history of trauma. No signs of rickets | — | Bilateral, less marked on left | Weakness of grasp. Difficulty in writing | — | Unusual prominence of the styloid process of the right ulna. Wrist and hand seem to be displaced backward on the forearm | — | Vibration to flexor muscles of the forearm | Restoration of muscular power. No change in degree of deformity |
| 68. | Robinson et Jacoulet 1909 | 34 F. | Nurse | 3 | No rickets. Fall on left hand at age 2 | — | Left | Muscular weakness No pain | Extension, pronation and supination diminished. Flexion increased past right-angle | Left arm shorter than right. Swelling at the back of the wrist due to head of ulna which is luxated backward from the radius and carpus. Anterior curvature of the radius and apparent anterior displacement of the hand. Roentgenogram | — | — | — |
| 69. | Robinson et Jacoulet 1909 | 27 F. | — | ? | — | — | Left | *Cadaveric Dissection* | | Ulna: Posterior projection of the head at the back of the wrist. Radius: Anterior curvature | — | *Cadaveric Dissection* | |
| 70. | MacLennan 1909 | 16 F. | — | 13 | Unassociated with stress or strain | — | Bilateral | Slight ache after heavy work | Decreased extension, balanced by an increase in flexion | Ulna: Lower end prominent. Radius: Curved laterally. Both forearms comparatively shortened | — | Cuneiform osteotomy | Good |
| 71. | Palazzi 1909 | 23 F. | Farmhand | 3 | No history of trauma. Hard work | — | Bilateral, especially the right | At onset, pain on hard work; then continual | Extension and abduction nil. Adduction diminished. Flexion increased | Apparent anterior dislocation at wrists. Dislocation at the lower radio-ulnar joint; reducible but returns on release of pressure. Radius: Bowed anteriorly. Left arm shortened. Roentgenogram | Left leg shorter than right. Right cubitus valgus | Osteotomy of the radius | Very satisfactory |
| 72. | Putti 1909 | Adult F. | — | Congenital | — | — | Bilateral | *Museum Dissection Specimen.* Discovered in Institute of Pathologic Anatomy, University of Bologne | | (Plaster model): Prominence of ulnar head at back of wrist. Anterior curvature of the radius and anterior displacement of the carpus and hand. Roentgenogram | *Museum Dissection Specimen* | | |
| 73. | Gaudier 1909 | 14 F. | Schoolgirl | 13 | No history of trauma or of hard work | — | Bilateral | Wrist movements are clumsy and slightly painful | Movements clumsy | Shortening of both forearms. Ulna: Head palpable below palm of hand, dislocated anteriorly from the radius. Radius: Bowed laterally with a slight posterior curvature. There is "pyramidalization" of the carpal bones. Roentgenogram | Clubfoot, left. Compensatory pelvic deformity and scoliosis. Bilateral genu valgum and genu recurvatum | Luxation of ulna is reducible | Deformity recurs on release of pressure |
| 74. | Ewald 1909 | 24 F. | Masseuse | Youth | No history of trauma | — | Bilateral | No pain | Extension limited 30° Supination impossible | Dislocation of the ulna, overriding the carpus superiorly. Hand and arm show a bayonet-shaped deformity. "Pyramidalization" of the carpal bones. Slight anterior bowing of the radius with normal relation to carpus. Roentgenogram | — | — | — |
| 75. | Gaugele 1909 | 15 F. | Governess | 15 | Fell from wagon at age 9. No trauma to wrist | — | Right | Pain in right wrist on washing | All motions limited. Extension, supination and pronation markedly decreased | Ulna: Slightly bowed; styloid process projects dorsally. Radius: Bowed anteriorly. Roentgenogram | — | — | — |
| 76. | Gaugele 1909 | 17 F. | Seamstress | Infancy | No rickets. Carried heavy weights | — | Bilateral | Pain short time | No limitation | Prominence of the ulnar head due to backward dislocation, reducible. Moderate anterior bowing of both radii. Roentgenogram | — | — | — |
| 77. | Gaugele 1909 | 19 M. | Weaver | Childhood | No rickets. No history of trauma | Father had similar deformities | Bilateral, mostly on left | Pain in wrists, especially at work | Motion normal on both sides | Prominence of the ulnar heads. Dislocation of the inferior radio-ulnar joints. Hand and carpus: Displaced anteriorly, in normal relation to radius. Articular end of radius: Deviated anteriorly and inward. Roentgenogram | — | — | — |
| 78. | Mauclaire et Labadie-Lagrave 1909 | — — | — | 2 | History of trauma | — | — | — | — | Luxation of the inferior ulnar articulation from the radius and carpus. Radius: Anterior bowing, but normal carpal articulation retained. Roentgenogram | — | — | — |
| 79. | Burnier et Neveux 1909 | 29 F. | Seamstress | 3 | Fall on wrist at age 3. No rickets or tuberculosis | — | Bilateral | Weakness of both hands. No pain | Flexion 90°; extension very limited | Ulna: Head has lost all connection with the wrist joint. Radius: Inferior extremity curves forward, taking the carpus and hand with it. There is no true luxation of the wrist. Roentgenogram | Subject to rheumatism and sciatica | Operative intervention refused | — |
| 80. | Leriche 1909 | 15 M. | Hatter | 14 | Fracture of the right ulna in infancy | — | Bilateral, especially the right | Pain on working relieved by rest | No limitations | Ulna: "Enormous swelling" at the back of the wrist, due to backward dislocation of ulnar head. Right radius: Bowed anteriorly. The hand appears luxated forward, but retains normal relationship with radius. Roentgenogram | — | Osteotomy of the right radius proposed | — |
| 81. | Stokes 1910 | 14 F. | Farmer | 12 | "Overstrained" both wrists at age 9 | — | Bilateral | Pain in wrist on work or exercise | Limitation of extension; otherwise normal | Ulna: Marked prominence of the styloid process. Radius: Markedly bowed forward in lower third. Roentgenogram | — | — | — |
| 82. | Stokes 1910 | 15 F. | — | — | No evidence of rickets | — | Bilateral | Pain on continuous use of wrist | Extension slightly limited | Ulna: Prominent styloid process at the back of the wrist, which overrides the carpus. Roentgenogram | Bowing of the tibiae | — | — |
| 83. | Jacoulet 1910 | 60 M. | Soldier, then carpenter | 18 | Fractured left radius after onset of deformity. No trace of rickets. Hard work | — | Bilateral, more marked on the right | No pain; no weakness | No limitations | Ulna: Prominent head; with luxation of the inferior radio-ulnar joint. Radius: Anterior curvature, accentuated at the metaphysis. Apparent anterior subluxation of the hand, but the relation of the carpus to radius remains normal. Roentgenogram | Atrophic cirrhosis. Pulmonary tuberculosis | Osteotomy not deemed advisable | Course: 7 yrs. (Progress of deformity ceased at age 25) |
| 84. | Jones 1911 | 60 F. | Housewife | 12 | No history of trauma, rickets or tuberculosis | — | Bilateral | Slight attacks of pain and weakness in both wrists | Flexion and abduction slightly diminished | Ulnae: Styloid processes both very prominent at the back of the wrist. Well-marked, characteristic deformity of the wrist. Roentgenogram | Sarcoma of the elbow | Plaster encasement | — |
| 85. | Springer 1911 | 12 F. | — | 5 | No history of trauma. No sign of rickets | — | Left | No pain at onset; later, pain severe | Extension and adduction markedly decreased. Flexion and abduction increased. Supination slightly decreased | Ulna: Prominence of the head at the back of the wrist. Radius: Anterior bowing, especially at the epiphysis. Ulna overrides the carpus. Deviation of the articular surface of the radius. Roentgenogram | Tuberculosis of the hip | See ref. 218 for operative interference, performed in 1913 | |
| 86. | Springer 1911 | 11 F. | — | — | Blow to wrist 14 days previous | — | Right | Pain in right wrist on motion | — | Slight deformity of right hand. Radius: Slight anterior bowing at the region of the epiphysis and moderate lateral bowing of the diaphysis. Radio-ulnar dislocation. Hemiatrophy of the ulnar side of the radial epiphysis. Roentgenogram | — | See ref. 218 for operative interference, performed in 1913 | |
| 87. | Springer 1911 | 14 F. | — | 12 | No evidence of rickets | | Bilateral, mostly right | No pain | — | Radius: Marked anterior and lateral bowing. "Pyramidalization" of the carpal bones. Carpus and hand: Displaced anteriorly. Roentgenogram | Flat-chested. Hyperostosis of the ulnar styloid | See ref. 218 for operative interference, performed in 1913 | |
| 88. | Codet-Boisse 1911 | 16 F. | Laundryironer | 13 | Hard work. No evidence of rickets | — | Bilateral, especially the right | Pain, increased at work. Tender to pressure | Extension limited to 35°. Pronation and supination limited | Ulnae: Prominence of the heads. Radius: Bowed anteriorly, and toward ulna. Ulna dislocated from carpus. Roentgenogram | — | Rest | Pain decreased |
| 89. | Brandes 1911 | aged M. | Baker, then wood-cutter, then writer | 18 | No sign of rickets | — | Bilateral | Pain at work; later continuous | Extension and abduction decreased. Pronation and supination limited | Bayonet-shaped deformity of the hands. Ulnae: Marked prominence of the heads. Radii: Anterior bowing. Roentgenogram | — | — | — |
| 90. | Brandes 1911 | 14 F. | Domestic servant | 11 | No sign of rickets | Daughter of above | Bilateral | Pain in wrists during late stage | Extension decreased 50%. Abduction impossible. Supination and pronation decreased | Ulnae: Prominence of distal ends. Shortening of both forearms. Radii: Anterior bowing, especially near the wrist. Roentgenogram | Small chest | Heat and massage. Bandage to prevent progression of deformity | Pain disappeared |
| 91. | Brandes 1911 | 8 F. | — | 6 | No sign of rickets | Sister of above | Bilateral | No pain | — | Deformity similar to older sister's above, but not so marked. Roentgenogram | — | — | — |
| 92. | Quadrone 1911 | 28 M. | Farmer | Birth | No history of trauma. No rickets | — | Bilateral | No pain, even on hard work | Slight limitation of flexion, extension, and abduction | Ulnae: Symmetrical retroposition of the heads, with an apparent subluxation forward of the hands at the wrists. Radii: Lower ends bent forward and downward. Roentgenogram | — | — | — |
| 93. | Ramos 1911 | 18 F. | Wealthy family | 8 | No history of trauma. Plays piano | — | Bilateral | — | — | Ulnae: Prominence of the heads. Radii: Anterior curvature, most marked in their lower thirds. Roentgenogram | — | — | — |
| 94. | Melchior 1912 | 19 F. | Farmhand | — | No sign of rickets | — | Bilateral | No pain | Flexion: R 60°; L. 45°. Extension: R. 65°; L. 70°. Pronation and supination normal | Carpus: Displaced laterally (marked cubitus valgus). Both hands in "fourchette" position. Ulnae: Prominence of the lower ends. Radii: Lower ends bowed anteriorly, more marked on the right. Roentgenogram | Shortening of fourth and fifth metacarpals (brachydactylia) of both hands | — | — |
| 95. | Magnus 1912 | 15 F. | — | 13 | No sign of rickets | Mother has deformity more marked | — | Pain in left wrist on hard work | Extension impossible. Abduction limited. Adduction normal | Bayonet-shaped deformity of the hand. Radius: Bowed anteriorly, and shortened. Ulna: Disarticulated from carpus and radius, and overrides the carpus dorsally. Roentgenogram | — | — | — |
| 96. | Taylor 1912 | 13 F. | Schoolgirl | 11 | No history of trauma. No sign of rickets | — | Bilateral | Pain in wrists, especially on working | Extension markedly restricted. Pronation and supination limited | Ulna: Heads very prominent at the back of the wrist. Hand: Displaced anteriorly. Roentgenogram | — | Two stage cuneiform osteotomy. Plaster of paris splint | Free from pain or deformity. Excellent function |
| 97. | Taylor 1912 | 15 F. | Schoolgirl | 13 | No trauma. No sign of rickets | — | Bilateral | Weakness. Pain over ulnar head | Motion nearly normal | Marked dorsal prominence of the lower end of the ulna | Slight scoliosis to the left | — | — |
| 98. | Berg 1913 | 19 F. | Servant | 17–18 | No sign of rickets | — | Left | Tenderness on palpation. Pain on hard work | All movements decreased | Left wrist thickened. Radius: Shortening, with slight bowing at the site of the epiphysis. Slight anterior displacement of the hand. Roentgenogram | — | Attempt at reduction and application of plaster encasement | Deformity found increased after 6 mos. |
| 99. | Berg 1913 | 16 F. | Peasant | 14 | History of hard work. No sign of rickets | — | Right | Painful at onset and during hard work | Extension and abduction decreased. Flexion increased. Adduction and supination normal | Ulna: Prominence of head. Hand and carpus: Displaced anteriorly, but retains normal relation with the articular surface of the radius. Radius: Bowed anteriorly. Luxation of the inferior radio-ulnar joint. Roentgenogram | — | Two attempts at reduction | No improvement |
| 100. | Finzi 1913 | 18 F. | — | | | — | Left | — | — | Radius: Obliteration of [illegible] | Exostosis [illegible] | | |

| | | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | | | | but not so marked. Roentgenogram | | | | |
| 92. Quadrone 1911 | 28 M. | Farmer | Birth | No history of trauma. No rickets | — | Bilateral | No pain, even on hard work | Slight limitation of flexion, extension, and abduction | Ulnae: Symmetrical retroposition of the heads, with an apparent subluxation forward of the hands at the wrists. Radii: Lower ends bent forward and downward. Roentgenogram | — | — | — | |
| 93. Ramos 1911 | 18 F. | Wealthy family | 8 | No history of trauma. Plays piano | — | Bilateral | — | — | Ulnae: Prominence of the heads. Radii: Anterior curvature, most marked in their lower thirds. Roentgenogram | — | — | — | |
| 94. Melchior 1912 | 19 F. | Farmhand | — | No sign of rickets | — | Bilateral | No pain | Flexion: R 60°; L. 45°. Extension: R. 65°; L. 70°. Pronation and supination normal | Carpus: Displaced laterally (marked cubitus valgus). Both hands in "fourchette" position. Ulnae: Prominence of the lower ends. Radii: Lower ends bowed anteriorly, more marked on the right. Roentgenogram | Shortening of fourth and fifth metacarpals (brachydactylia) of both hands | — | — | |
| 95. Magnus 1912 | 15 F. | — | 13 | No sign of rickets | Mother has deformity more marked | — | Pain in left wrist on hard work | Extension impossible. Abduction limited. Adduction normal | Bayonet-shaped deformity of the hand. Radius: Bowed anteriorly, and shortened. Ulna: Disarticulated from carpus and radius, and overrides the carpus dorsally. Roentgenogram | — | — | — | |
| 96. Taylor 1912 | 13 F. | Schoolgirl | 11 | No history of trauma. No sign of rickets | — | Bilateral | Pain in wrists, especially on working | Extension markedly restricted. Pronation and supination limited | Ulna: Heads very prominent at the back of the wrist. Hand: Displaced anteriorly. Roentgenogram | — | Two stage cuneiform osteotomy. Plaster of paris splint | Free from pain or deformity. Excellent function | |
| 97. Taylor 1912 | 15 F. | Schoolgirl | 13 | No trauma. No sign of rickets | — | Bilateral | Weakness. Pain over ulnar head | Motion nearly normal | Marked dorsal prominence of the lower end of the ulna | Slight scoliosis to the left | — | — | |
| 98. Berg 1913 | 19 F. | Servant | 17–18 | No sign of rickets | — | Left | Tenderness on palpation. Pain on hard work | All movements decreased | Left wrist thickened. Radius: Shortening, with slight bowing at the site of the epiphysis. Slight anterior displacement of the hand. Roentgenogram | — | Attempt at reduction and application of plaster encasement | Deformity found increased after 6 mos. | |
| 99. Berg 1913 | 16 F. | Peasant | 14 | History of hard work. No sign of rickets | — | Right | Painful at onset and during hard work | Extension and abduction decreased. Flexion increased. Adduction and supination normal | Ulna: Prominence of head. Hand and carpus: Displaced anteriorly, but retains normal relation with the articular surface of the radius. Radius: Bowed anteriorly. Luxation of the inferior radio-ulnar joint. Roentgenogram | — | Two attempts at reduction | No improvement | |
| 100. Pinzi 1913 | 18 F. | — | — | — | — | Left | — | — | Radius: Obliteration of inner third of radial epiphysis; rotation of joint surface down and inward. Ulna: Longer than radius, with subluxation of carpo-ulnar joint. Roentgenogram | Exostosis of left radius at epiphyseal line | — | — | |
| 101. Adler 1914 | — F. | — | — | No trauma. No sign of rickets | — | Bilateral | No pain | Slight restriction of extension | Radius and ulna: Marked prominence posteriorly, especially the ulna. The wrists appear thick. Roentgenogram | — | — | — | |
| 102. Parkes 1915 | 15 F. | — | 12 | — | — | Right | Pain and tenderness of right wrist | Inability to extend wrist. Flexion more marked than normal | Ulna: Lower end projects posteriorly. Roentgenogram | — | Cuneiform osteotomy and Lane plate | Deformity corrected. No pain | |
| 103. Earl 1916 | 19 F. | — | 17 | — | — | Bilateral | Pain in both wrists | — | Marked deformity of both wrists. General relaxation of all joint structures. Roentgenogram | Amenorrhea. Infantile uterus | — | — | |
| 104. Mauclaire 1916 | 38 | — | 10 | No trauma. No sign of rickets | — | Bilateral | Slight pain | Limitation of flexion and extension | Ulna: Head luxated backward from radius and carpus. Radius: Bowed forward carrying the carpus anteriorly and to the ulnar side | — | — | — | |
| 105. Mauclaire 1916 | 17 M. | — | — | Plays a lot of tennis | — | Bilateral | — | — | Ulna: Slight luxation of the head from the radius and wrist. Radius: Projection of the epiphysis forward. Hand and wrist: In normal relation to radius | — | — | — | |
| 106. Mauclaire 1916 | 16 F. | — | 15 | Plays violin. No sign of rickets | — | Bilateral, especially the right | — | — | Ulna: Posterior luxation. Radius: Lateral curvature. The epiphyseal line is irregular on the right, missing on the left. Roentgenogram | — | — | — | |
| 107. Guye 1919 | 17 F. | — | 11 | Walked late. Frontal bossing. No other sign of rickets | — | Bilateral, more marked on left | No pain | Hypermotility of flexion and extension | Ulna: Luxation of the inferior articulation with prominence of the head at the back of the wrist. Radius: Anterior bowing. Apparent anterior subluxation of the hand. "Pyramidalization" of the carpal bones. Roentgenogram | — | — | — | |
| 108. Pillate 1919 | 35 F. | Dairymaid | 6 | Fall on hands at age 6. No rickets | — | Bilateral, especially the left | No pain. Able to do hard work | — | Ulna: Prominence of the head at back of wrist. Radius: Lateral curvature of the shaft and an anterior curvature of the lower third. "Pyramidalization" of the carpal bones. Roentgenogram | Bilateral cubitus valgus | — | — | |
| 109. Pillate 1919 | — F. | Housewife | 11 | No trauma. No rickets | Newborn child has absence of radius | Bilateral, especially the right | Easily fatigued. Pain from onset, even on light work | Flexion limited; extension, pronation and supination are normal | Ulna: Marked prominence of the head, showing posterior dislocation. Radius: Curved anteriorly and somewhat laterally. Carpus: In normal relationship to radial articulation. "Pyramidalization" of the bones of the wrist. Roentgenogram | — | — | — | |
| 110. Pillate 1919 | 43 F. | Bottler | 11 | No trauma. No rickets | — | Bilateral | Pain during the first year only | Pronation and supination normal. Extension limited. Flexion exaggerated | Marked swelling at back of wrist at site of the ulnar head. Both forearms shortened. Radii: Bowed laterally and anteriorly. Hemiatrophy of ulnar half of radial epiphysis. Roentgenogram | — | — | — | |
| 111. Gery et Colombier 1920 | 70 F. | Cook | 30 (?) | No trauma or hard work. Kneads dough | — | Bilateral | — | Flexion and pronation diminished. Abduction practically nil | Ulna: Luxated lower end. Radius: Anterior curvature of the lower end, with moderate lateral curvature of the shaft. "Pyramidalization" of the bones of the carpus | Patient died of gastric carcinoma | *Cadaveric Dissection* | | |
| 112. Gery et Colombier 1920 | 62 F. | Seamstress | 3 | No rickets | — | Bilateral | Slight pain | Little functional limitation | Ulna: Luxation of head, reducible on pressure. Radius: Slight bowing laterally, and anteriorly at the level of the epiphysis | — | — | — | |
| 113. Wéry 1921 | — F. | — | 14 | — | Sister also has deformity | Bilateral | Pain first 2 yrs. | — | Radius: Double curvature of lower end anteriorly and laterally. Luxation of the inferior radio-ulnar joint and a pseudo-luxation of the hand and carpus. Roentgenogram | — | — | — | |
| 114. Wéry 1921 | — F. | — | 14 | — | — | Bilateral | Pain during first 2 yrs. | — | Radius: Lower end bowed anteriorly and laterally. Ulna: Dislocated from radius. Apparent forward luxation of hand and carpus. Roentgenogram | — | — | — | |
| 115. Masmonteil 1921 | 17 M. | Student | 14 | No history of trauma | — | Left | Pain | Extension, abduction, and adduction zero. Pronation 90°; flexion 15°; supination 45° | Prominence of the head of the ulna. Palmar subluxation of the hand | — | Cuneiform osteotomy. Hand immobilized in hyperextension in a plaster encasement. Later resection head of ulna | Esthetically perfect. Pronation 180°; extension 80°; flexion 22°; abduction 20°; adduction 25° | |
| 116. Mathieu et Joseph 1922 | 16 F. | Tile-maker | 15 | Carries heavy weights | — | Bilateral, especially the left | Left wrist is painful | All movements normal in amplitude | Hand and carpus: Apparent dislocation anteriorly. Radius: Bowed anteriorly and laterally, with a thick exostosis about the middle on the ulnar side. Luxation of the inferior radio-ulnar articulation. Roentgenogram | Bilateral bowing of the lower third of the tibiae | Deformity reducible, but returns on release of pressure. Operation refused | — | |
| 117. Mathieu et Joseph 1922 | 16 F. | Scholar | 13 | Does much writing | — | Right | Pain in right wrist, especially on writing | Extension limited; all other movements are normal | Swelling ulnar side of dorsum of wrist. Luxation of the inferior radio-ulnar joint. Radius: Anterior curvature at the metaphysis. Hand and carpus: In normal relation to radius. Roentgenogram | — | Ulna shortened by osteotomy | Presumed that functional result will be excellent | |
| 118. Brown 1923 | 11 F. | — | 8 | No evidence of rickets | — | Bilateral | Pain in wrist, especially on motion | Extension and abduction limited | Radii: Bowed with convexity posterior. Wrist: Apparently displaced forward. Roentgenogram | Bowing of both tibiae | — | Progressive | |
| 119. Greig 1924 | — F. | — | Early childhood | — | — | Bilateral | No pain | No limitation of motion | Ulna: Distal end overlaps carpus dorsally. Apparent disjunction of the head of the ulna from radius and wrist joint. Forearm unusually short. Radius: Curved anteriorly. Roentgenogram | — | — | — | |
| 120. Ianni 1924 | 26 F. | — | 11–12 | No trauma. No hard work | — | Left | Pain on playing piano. Tires on movement | Extension limited. Abduction, supination and pronation slightly limited. Adduction increased. Flexion normal | Hand: Apparent forward dislocation. Ulna: Overrides the hand and causes a swelling at the back of the wrist. Radius: Anterior curvature of the lower third. Roentgenogram | — | Transverse linear osteotomy of the radius at age 13 | Deformity cured and perfect function at wrist one month after operation | |

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TABULATION OF ANALYSES OF RELEVANT DATA OF 171 CASES OF TRUE MADELUNG'S DEFORMITY (*Continued*)

| No. | Author and Year | Age and Sex | Occupation | Age at Onset | Casual Relationship | Heredity | Unilateral or Bilateral | Wrist Joint: Symptoms | Wrist Joint: Function | Examination | Other Abnormalities | Treatment | Course or Result |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 121. | Levyn 1924 | 56 F. | Housewife | 12 | No history of trauma or rickets | Mother of patient below | Bilateral | No pain, but wrists are fatigued more easily than normal | Extension limited. Flexion increased | Shortening of radius and ulna with slight bowing of both bones. Pronounced backward dislocation of ulna. Excavated, wedge-shaped appearance of articular surfaces of radius and ulna. Roentgenogram | — | Glandular therapy (thyroid, pituitary and antuitrin) | — |
| 122. | Levyn 1924 | 18 F. | Student | 12 | No rickets or trauma. Played piano a lot | Daughter of above | Bilateral | No pain. Slight exertion produces fatigue | Extension limited. Flexion increased | Bowing of radius not so marked as above. Excavated and wedge-shaped articular surfaces. Roentgenogram | — | Glandular therapy (thyroid, pituitary and antuitrin) | Increase in length of ulna. No change in deformity |
| 123. | Moore 1924 | 16 F. | Typist; plays piano | 14 | No history of trauma or hard work | — | Left | Pain in wrist | Extension limited. Flexion unusually free | Left hand. Displaced forward. Ulna: Head very prominent. Radius: Lower fourth bowed forward. Roentgenogram | — | Cuneiform osteotomy. Plaster encasement, in extension | Deformity corrected; extension increased; flexion now normal |
| 124. | Moore 1924 | 15 M. | — | 13 | Patient boxed a great deal | — | Left | Pain in wrist | Extension limited. Flexion unusually free | Ulna: Marked backward projection of the head. Radius: Some forward curvature. Hand: Deviated to ulnar side. Roentgenogram | — | Resection of the head of the ulna. Sutured lower end of ulna to carpus | Good |
| 125. | Define 1924 | 13½ F. | Weaver; plus housework | 12 | Hard, exhausting Work. No trauma or rickets. Positive Wassermann | Luetic | Bilateral | Intermittent pain in wrists and forearms, aggravated by motion. Weakness of wrists | Extension diminished and painful. Supination and adduction decreased. Pronation and flexion normal | Swelling at the back of the wrist with apparent anterior displacement of the hand. Radius: Anterior curvature, with atrophy of the ulnar side of the epiphysis. Inferior ulnar articulation luxated from radius and carpus. Roentgenogram | — | Massage, active and passive motion. Osteotomy considered and will be performed at proper time | — |
| 126. | Garrido-Lestache 1925 | 13 M. | Field-hand | 11 | — | — | Right | No severe pain. Weakness of right wrist | Extension limited. Flexion augmented | Apparent anterior displacement of hand. Swelling of ulnar head at back of wrist. Radius: Anterior bowing. Roentgenogram | — | Immobilization and rest | — |
| 127. | Llardo and Gallardo 1925 | 14 F. | — | 13 | — | — | Bilateral, especially the right | Fatigue in the right wrist | Extension, adduction, and supination limited on right side | Carpus: Apparent anterior luxation. Ulna: Prominence of the head at the back of the wrist. Radii: Anterior bowing. Roentgenogram | — | — | — |
| 128. | Merlini 1925 | 25 F. | Housework | 11 | No history of trauma. No evidence of rickets | — | Bilateral | Continuous, vague pain in the wrists during course of disease | Extension and abduction reduced. Pronation and supination not modified. Flexion and adduction more thanf normal | Ulnae: Marked prominence of heads. Forearms: Shortened. Radii: Anterior and lateral bowing of shafts. Dehiscence of the inferior radio-ulnar articulation. Hand and carpus: Displaced anteriorly, in normal relation to the articulating surface of the radius. Roentgenogram | — | — | — |
| 129. | Mazzini 1925 | 17 F. | Servant | 7 | No evidence of rickets. | — | Bilateral, especially the right | Pain on hard work, relieved by rest | Left side normal. Right: Abduction and adduction restricted. Extension zero | Ulna: Prominence of the head. Forearms: Marked shortening. Radius: Anterior bowing. Roentgenogram | — | — | — |
| 130. | DeBernardi 1925 | 10 F. | — | 4 | No history of trauma | — | Left | Pain at onset and on palpation | Flexion increased. Extension limited | Left forearm shortened, deformity at the back of the wrist. Luxation of the inferior radio-ulnar joint. Epiphysis of the radius turned anteriorly. Hand displaced forward, in normal relation to the articulating surface of the radius. Roentgenogram | — | — | Course: 18 mos. |
| 131. | DeBernardi 1925 | 8 F. | Schoolgirl | 3 | No trauma. No evidence of rickets | — | Right | Pain at onset. Stabbing pain on motion | Extension nil. Abduction limited | Swelling at the back of the wrist. Radius. Bowed anteriorly at the region of the epiphysis. Hand: Displaced anteriorly. Roentgenogram | — | — | — |
| 132. | Jòsa 1926 | 17 M. | Apprentice blacksmith | 14 | Hard work | Orphan; no recollection of family | Bilateral, especially the left | Slight pain at onset relieved by work | Extension: R. 48°; L. 35°. Flexion: R. 78°; L. 81°. Adduction: R. 40°; L. 26°. Abduction: R. 10°; L. 26°. Supination normal | Hands: Bayonet-shaped deformity. Ulnae: Prominence of the heads. Radius: Shorter than ulna, more marked on the left side. Roentgenogram | Scoliosis and genu valgum. Inguinal hernia | — | — |
| 133. | Catterina 1926 | 33 F. | Servant | Childhood | — | — | Bilateral | No pain in the wrists | Extension limited. Flexion and adduction increased | Radius: Marked anterior and lateral curvature of the lower third, with moderate curvature of the upper two-thirds. Ulna: Slightly bowed forward, heads prominent at back of wrist. Hand: Displaced forward. Roentgenogram | — | — | — |
| 134. | Pedrazzi 1927 | 12 F. | Schoolgirl | 8 | No trauma. Possible rickets | Mother and younger sister have similar deformities | Bilateral, especially the left | Pain on grasping, writing, sewing, and hard work | Extension difficult. Flexion easy. Marked limitation of abduction and adduction | Swelling at the back of the left wrist. Apparent anterior dislocation of the wrists. Radii: Bowed anteriorly. Left arm shortened. Roentgenogram | Patient quite small in size for her age | — | — |
| 135. | Pedrazzi 1927 | 15 F. | — | 11 | Possible rickets. Plays piano. No trauma | Sister of above | Bilateral, especially the right | More or less continual pain | Flexion easy. Extension difficult. Adduction and abduction about normal | Ulna: Styloid prominent on right side. Radius: Curved laterally and anteriorly. Hand: Displaced forward. Roentgenogram | Patient quite small in size for her age | — | — |
| 136. | Foschini 1927 | 20 M. | Shoemaker | 11 | No history of trauma. No sign of rickets | — | Bilateral, especially the left | Pain on working | — | Swelling on ulnar side of back of wrists. Dislocation of the inferior radio-ulnar joint. Radius: Anterior curvature, most marked in the lower third. "Fork" hand deformity, with anterior displacement of the hand. Roentgenogram | — | — | — |
| 137. | Tollas 1927 | 16 F. | — | 11 | — | — | Bilateral, right more than left | Pain from onset, with gradual increase during progression of the deformity | Extension: R. 45°; L. 50°. Flexion: R. 90°; L. 90°. On both sides: Pronation normal; supination decreased; abduction 45°; adduction 10° | Ulna: Prominence of the heads. Carpus: Apparent anterior luxation. Bilateral cubitus valgus, right: 35°, left: 30°. Roentgenogram | — | — | — |
| 138. | Roederer 1928 | 30 F. | Domestic servant | Puberty | — | — | Bilateral | Weakness, clumsiness; unable to do hard work | Extension slightly limited. Flexion and pronation normal | Ulna: Abnormal swelling of the head above the wrist; reducible on pressure. Subluxation of the ulna from the radius and carpus. Radius: Anterior bowing at the metaphysis. Roentgenogram | Patient very small in size | Deformity reduced by pressure, and held in place by wrist band | Relief of pain for 3 mos. |
| 139. | Bertoletti 1928 | 23 F. | Peasant | 18–19 | Hard work. No evidences of rickets | — | Bilateral, especially the right | Pain on motion and change of weather | Limitation of extension and abduction. Increase of flexion and adduction | Ulna: Bilateral prominence of the head, especially on the right. Radii: A moderate lateral and anterior curvature, bringing the articular surfaces down and inward. Hands: Displaced forward | — | Operative interference not deemed justifiable | Course: 4 yrs. Deformity still present, pain less |
| 140. | Félix 1928 | 16 F. | Handworker | 15½ | No history of trauma. No rickets | — | Bilateral | Pain in both wrists | Extension 20°; flexion 80°–90°; supination decreased; adduction less | Bayonet-shaped deformity of wrist. Ulna: Prominence of the heads. Radii: Marked anterior bowing; the ulnar third of the lower epiphysis is missing. Roentgenogram | Exostosis of both ulnae near epiphyseal line | — | — |
| 141. | Schnek 1928 | 18 M. | — | 10 | No history of trauma. No rickets | — | Right | Right hand weak. No pain | Right hand: Extention and abduction nil | Right forearm shorter than left. Carpus: Subluxated anteriorly. Radius: 5 cm. shorter than ulna. Roentgenogram | Small in stature | Triangular osteotomy of the radius. Resection of the ulnar head. Plaster encasement, with hand fixed in extension | Three mos. later: Good function and all movements present |
| 142. | Schnek 1928 | 36 F. | — | Youth | Fracture of radius at age 2; badly set | — | Bilateral | Pain on making a fist | Flexion 85°; extension 20°; abduction 25°; adduction none | Radii: Anterior bowing. Forearms: Shortened. Carpus: Subluxated; more marked on right | — | Osteotomy and resection as above | — |
| 143. | Vidal 1929 | 45 F. | Servant | Infancy | Hard work. No trauma | Mother, two sisters and a niece have similar deformities | Bilateral | No pain. Able to do hard work | Flexion 80°–90°; extension 45°; abduction limited; adduction augmented. Pronation and supination normal | Ulna: Prominence of the head. Dislocation of the inferior radio-ulnar joint. Hand and carpus: Apparent anterior displacement. Radius: Lower fourth is curved laterally and anteriorly. Roentgenogram | Bilateral, pulmonary tuberculosis | — | — |
| 144. | Vidal 1929 | 50 F. | Cook | Infancy | No history | Sister of above | Bilateral | Pain on exaggerated motion of wrists | Extension 45°. Flexion 80°–90°. Abduction limited. Adduction increased. Pronation and supination normal | Ulna: Head projects over the carpus. Radius: Lower third curved anteriorly. Hand and carpus: Displaced forward, in normal relationship to the radius. Roentgenogram | — | — | — |
| 145. | Fazio 1930 | 14 F. | — | 12 | No history of trauma. No rickets | — | Bilateral, especially the right | Pain at onset | Active motion present | Radius: Slight anterior curvature at the region of the epiphysis, and slight lateral curvature of the shaft. Hand and carpus: Anterior displacement. Roentgenogram | Slight of stature | Oblique linear osteotomy. Application of plaster encasement | "Cured" |
| 146. | Rocher et Roudil 1930 | 24 M. | — | 7 | No history of trauma | — | Bilateral | No trouble except on hard work | Hands held in fixed pronation. Supination impossible. Extension and flexion normal | Ulnae: Prominence of the heads, due to luxation from the inferior end of the radius. Radius: Lateral bowing of the diaphysis and anterior bending at the epiphysis. Hemiatrophy of the epiphysis on the ulnar side. Roentgeno gram | — | — | — |
| 147. | Cserey-Pechany 1930 | 18 F. | — | 12 | Irregular menses. No evidences of rickets | — | Bilateral | — | — | Dislocation of the inferior radio-ulnar articulations. Radii: Bowed anteriorly and laterally. Hemiatrophy of the inner half of the inferior radial epiphysis. Roentgenogram | — | Ovarian and glandular hormone therapy for 7 mos. | Course ceased. Roentgenogram "evidence" of improvement of the deformity |
| 148. | Gazzotti 1931 | 18 F. | Ironer (presser) | 14 | No trauma. No rickets. | — | Bilateral | Slight pain on all movements | Extension and abduction reduced. Pronation and supination limited. Adduction and flexion increased over normal | Radii: Bilateral anterior and lateral bowing. Ulnae: Override the carpus superiorly. Hands: Displaced anteriorly in normal relation to radius. Roentgenogram | — | Oblique osteotomy (bilateral) and application of plaster encasement | Very good. Ten mos. later all movements present and deformity cured |
| 149. | Tancredi 1931 | 15 F. | — | 11 | Menses irregular | — | Bilateral | — | Supination impossible. Extension, abduction, adduction and pronation limited. Flexion increased | Radii: Anterior curvature, especially in the distal fourth. Ulnae: Heads project dorsally. Hands: Displaced anteriorly. Roentgenogram | Legs and arms are short compared to trunk | Transverse osteotomy and application of plaster encasement | Perfect result functionally and morphologically |
| 150. | Vianna 1931 | 13 F. | — | 11 | — | — | Bilateral | Pain—first during day, then continuous | Abduction, adduction, pronation and supination limited. Extension nil. Flexion increased | Wrist: Bayonet-shaped deformity, with prominence of the ulnar heads. Radius: Anterior bowing, especially in the lower third. Dislocation of the inferior radio-ulnar articulation. Roentgenogram | Spina bifida occulta of of the lower cervival and upper thoracic vertebrae | Transverse osteotomy with correction of deformity, first right, then left | "Brilliant" |
| 151. | Fick und Pahil 1931 | 21 F. | Servant | 19 | No rickets | — | Right (beginning on left ?) | Work produces pain and fatigue | Marked limitation of extension and flexion. Abduction normal | Ulna: Prominence of the distal end. Carpus: Anterior subluxation. Radius: Marked lateral and slight anterior bowing. Roentgenogram | — | — | — |
| 152. | Salisachs 1933 | 16 M. | Student | Infancy | — | — | Bilateral, especially the right | Slight pain over lower third of radius | Limitation of extension | Subluxation of the inferior radio-ulnar joint. Radius: Anterior curvature. Ulna: Head prominent. Roentgenogram | Hyperostosis of ulnar epiphysis | Immobilization in splint | — |
| 153. | Salisachs 1933 | 13 F. | — | Infancy | Hard work for 4 to 5 yrs. | Father has similar deformity | Bilateral, especially the right | No discomfort | Extension, pronation, and supination limited. Abduction impossible. Flexion exaggerated | Ulnae: Prominence of heads. Radii: Lateral curvature | Kyphosis | — | — |
| 154. | Salisachs 1933 | 25 M. | Writer | — | — | — | Bilateral, especially the right | No pain | Extension slightly limited. Contracture of flexors limits other movements | Hands: Apparent marked anterior displacement. Radii: Anterior and lateral bowing, most marked in the lower half. Roentgenogram | Cystic areas noted throughout the skeletal system | — | — |

| | | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 1930 | F. | — | 12 | Irregular menses. No evidences of rickets | — | Bilateral | — | — | Dislocation of the inferior radio-ulnar articulations. Radii: Bowed anteriorly and laterally. Hemiatrophy of the inner half of the inferior radial epiphysis. Roentgenogram | — | Ovarian and glandular hormone therapy for 7 mos. | Course ceased. Roentgenogram "evidence" of improvement of the deformity |
| 148. Gazzotti 1931 | 18 F. | Ironer (presser) | 14 | No trauma. No rickets. | — | Bilateral | Slight pain on all movements | Extension and abduction reduced. Pronation and supination limited. Adduction and flexion increased over normal | Radii: Bilateral anterior and lateral bowing. Ulnae: Override the carpus superiorly. Hands: Displaced anteriorly in normal relation to radius. Roentgenogram | — | Oblique osteotomy (bilateral) and application of plaster encasement | Very good. Ten mos. later all movements present and deformity cured |
| 149. Tancredi 1931 | 15 F. | — | 11 | Menses irregular | — | Bilateral | — | Supination impossible. Extension, abduction, adduction and pronation limited. Flexion increased | Radii: Anterior curvature, especially in the distal fourth. Ulnae: Heads project dorsally. Hands: Displaced anteriorly. Roentgenogram | Legs and arms are short compared to trunk | Transverse osteotomy and application of plaster encasement | Perfect result functionally and morphologically |
| 150. Vianna 1931 | 13 F. | — | 11 | — | — | Bilateral | Pain—first during day, then continuous | Abduction, adduction, pronation and supination limited. Extension nil. Flexion increased | Wrist: Bayonet-shaped deformity, with prominence of the ulnar heads. Radius: Anterior bowing, especially in the lower third. Dislocation of the inferior radio-ulnar articulation. Roentgenogram | Spina bifida occulta of of the lower cervival and upper thoracic vertebrae | Transverse osteotomy with correction of deformity, first right, then left | "Brilliant" |
| 151. Fick und Pahil 1931 | 21 F. | Servant | 19 | No rickets | — | Right (beginning on left ?) | Work produces pain and fatigue | Marked limitation of extension and flexion. Abduction normal | Ulna: Prominence of the distal end. Carpus: Anterior subluxation. Radius: Marked lateral and slight anterior bowing. Roentgenogram | — | — | — |
| 152. Salisachs 1933 | 16 M. | Student | Infancy | — | — | Bilateral, especially the right | Slight pain over lower third of radius | Limitation of extension | Subluxation of the inferior radio-ulnar joint. Radius: Anterior curvature. Ulna: Head prominent. Roentgenogram | Hyperostosis of ulnar epiphysis | Immobilization in splint | — |
| 153. Salisachs 1933 | 13 F. | — | Infancy | Hard work for 4 to 5 yrs. | Father has similar deformity | Bilateral, especially the right | No discomfort | Extension, pronation, and supination limited. Abduction impossible. Flexion exaggerated | Ulnae: Prominence of heads. Radii: Lateral curvature | Kyphosis | — | — |
| 154. Salisachs 1933 | 25 M. | Writer | — | — | — | Bilateral, especially the right | No pain | Extension slightly limited. Contracture of flexors limits other movements | Hands: Apparent marked anterior displacement. Radii: Anterior and lateral bowing, most marked in the lower half. Roentgenogram | Cystic areas noted throughout the skeletal system | — | — |
| 155. Kun 1933 | 14 F. | — | 13 | History of extreme effort, but after onset | — | Bilateral especially the left | Pain in both wrists | Extension and supination limited. Abduction nil. Flexion augmented | Ulnae: Posterior luxation of the inferior end. Radii: Anterior curvature of entire shafts, especially at the distal ends. "Pyramidalization" of the carpal bones. Roentgenogram | Spina bifida occulta fourth lumbar. Asymptomatic sacralization fifth lumbar vertebra. Pes valgus. Genu valgum | Plaster encasement applied, with hand in extension. Osteotomy considered for later date | Encasement removed because of pain. No improvement |
| 156. Massabuau, Solas et Nichet 1934 | — F. | — | 11 | No evidence of rickets | History of similar deformity in father | Bilateral | Pain on prolonged use of hand in writing | Diminished extension and supination | Ulna: Prominence of head. Radius: Anterior bowing. Carpus: Apparent anterior luxation. Roentgenogram | — | No operative interference | Course: 3 yrs. |
| 157. Kajon 1934 | 7 M. | — | 7 | No history of trauma. No rickets | — | Right | No pain | Extension slightly exaggerated. Abduction decreased | Right forearm: Shorter than left. Radius: Bowing, and widening of the epiphysis. Roentgenogram | Cervical rib | — | — |
| [illegible] et Rocher 1934 | 57 F. | — | Childhood | — | — | Bilateral, especially the right | No pain | No limitation of motion | Ulnae: Slight prominence of the heads at the back of the wrists. Dislocation of the inferior radio-ulnar joints. Radius: Slight anterior bowing of the radius at the epiphysis. Roentgenogram | Absence of the right caput humerus, and maldevelopment of the tuberosity | — | — |
| 159. Canton 1935 | 23 M. | — | Childhood | No history of trauma | — | Bilateral | No pain | Hands held in fixed pronation. Supination impossible. Extension and flexion normal | Ulna: Prominence of the head at the back of the wrists. Radius: Shaft bowed laterally, and anteriorly at the distal extremity. Roentgenogram | — | — | — |
| 160. Barsoum 1935 | 12 F. | — | 11 | — | — | Bilateral | Pain on extremes of motion. Weakness of wrist | "Abnormal" motility of both wrist joints | Hands and carpus: Symmetrical volar displacement. Radii: Abnormal curvature, with thickening at the distal end. Roentgenogram | — | — | — |
| 161. Beder und Heinismann 1935 | 25 F. | Factory worker | 12–13 | Amenorrhea. No development of breast | — | Bilateral | Pain in radiocarpal joint. General weakness | Limitation of extension | Ulna: Styloid prominent and projects dorsally. Radius: Bowed forward. Hand and carpus: Displaced anteriorly. Roentgenogram | Male configuration. Deformity upper tibial epiphysis | Suggests endocrine therapy | — |
| 162. Beder und Heinismann 1935 | 28 F. | — | 14 | Oligomenorrhea | — | Bilateral | — | — | Deformity of radiocarpal joint as above. Roentgenogram | — | — | — |
| 163. Landivar y Iparraguirre 1936 | 61 M. | Barrendero | 51 (?) | No evidence of rickets | — | Bilateral | No pain or discomfort | Extension 35°; flexion 90°. Adduction 50°; abduction 10°. Pronation and supination 30° | Radius: Anterior bowing. Separation of radio-ulnar articulation. Carpus: Anterior subluxation. Roentgenogram | Rarefied areas in left ileum, pubis and ischial tuberosity | — | — |
| 164. Chierici 1936 | 15 F. | — | 13½ | — | — | Bilateral | Weakness | Pronation, supination, and extension limited. Flexion exaggerated. Marked limitation of abduction; slight limitation of adduction | Ulnae: Prominence of heads at back of wrists. Radius: Anterior curvature in the lower third, most marked at the metaphysis. Lateral bowing of the lower fourth. Hand and carpus: Displaced forward, but in normal relation to the articular surface of the radius. Roentgenogram | — | — | — |
| 165. Chierici 1936 | 13 F. | — | 12 | Spontaneous | — | Bilateral | — | Extension and abduction limited. Adduction slightly limited. Flexion to 90° | Ulna: Abnormal swelling at the back of the wrist due to head of the ulna. Radius: Anterior curvature at the metaphysis. Hand and wrist: Anterior displacement, in normal relation to the radius. Roentgenogram | Curvature of both tibiae | — | — |
| 166. Claiborne and Kuntz | 16 F. | — | 9 | — | — | Bilateral, more marked on right | Pain in wrist, especially on motion | Extension and abduction limited | Ulnae: Heads prominent. Radii: Bowed (with the convexity) dorsally, and laterally. Carpus: Appears dislocated anteriorly. Roentgenogram | — | — | — |
| 167. Roudil, Drevon, et Mourgues 1936 | 14½ F. | — | 10½ | No stigmata of rickets. No history of trauma | — | Bilateral | Pain, especially on motion. Weakness of wrists | Supination 40°; pronation 70°; flexion 90°; extension normal; adduction 10°; abduction 30° | Ulna: Luxation of the inferior end from the radius and carpus. Forearms: Shortened. Radius: Lower third bowed anteriorly; moderate lateral bowing of the shaft. Hand and carpus: Displaced anteriorly, in normal relation to the radius. "Pyramidalization" of the carpal bones. Roentgenogram | Bilateral cubitus valgus | — | — |
| 168. Rocher 1937 | 13½ M. | — | 9 | No history of trauma. No rickets | — | Bilateral, more marked on right | "Growing" pain in wrists and knees | Flexion: R. 80°; L. 90°. Extension: R. 0°; L. 40°. Abduction: R. 0°; L. 20°. Adduction: R. 15°; L. 25°. Pronation normal. Supination normal on effort | Ulna: Projection of head at the back of the wrist. Radius: Marked anterior curvature, carrying the hand forward. No luxation of the wrist from the radius, but the radius is dislocated from the ulna. "Pyramidalization" of the carpal bones. Roentgenogram | Dischondroplasia of the os centrale of the wrist | — | — |
| 169. Burrows 1937 | 14 M. | Schoolboy and junior clerk | 8 | Sprained right wrist at age 2 | — | Right | — | Extension full. Abduction increased. Adduction negligible. Flexion limited to 50°. Supination limited | Ulna: Head of the ulna projects below the level of the radius. Characteristic silver-fork deformity of the "reverse" type of Madelung's. Hand and carpus: Displaced backward. Roentgenogram | — | Excision of the ulnar head. Linear osteotomy. Plaster encasement | Functional improvement |
| 170. Burrows 1937 | 17 M. | Warehouseman | 16 | — | — | Right | Pain, worse after work | — | Ulna: Lower end prominent at the back of the wrist. Radius: Bowed anteriorly. Hand: Displaced forward. Roentgenogram | — | Operation as above | Correction good. Motion normal. No shortening |
| 171. Anton, Reitz, and Spiegel 1938 | 18 F. | — | 15 | Hit by auto at age 9; no trauma to wrists | — | Bilateral | Pain in both wrists | All movements, especially extension and abduction, limited. (See case history for details) | Forearms: Shortened. Ulnae: Heads project at back of wrists. Radii: Bowed anteriorly at the region of the metaphysis, with moderate lateral curvature of the shaft. Hands: Displaced forward. "Pyramidalization" of the carpal bones. Roentgenogram | Increased digital impressions of the inner table of the cranial vault | Rest. To return for osteotomy | — |

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and B), as if the hands had been dislocated anteriorly. There is also an obvious shortening of both forearms (left—eight and one-quarter inches long; right—eight and one-half inches long). A lateral view of the forearm and hand roughly resembles a bayonet (Fig. 1 A). There is a marked swelling at the back of the wrist which palpation determined as being the head of the ulna and which, apparently, overrode the carpus superiorly. Palpation of the lower end of the radius reveals a definite forward bowing with convexity on the dorsal surface, and a moderate lateral bowing with the concavity toward the ulna. This bowing has displaced the carpus and hand forward and to the ulnar side. The flexor tendons are slightly more prominent than usual. Measurements of the forearms show:

| | Right | Left |
|---|---|---|
| Radius (from the head to the styloid process) | 18 cm. | 18½ cm. |
| Ulna (from olecranon to styloid process) | 20½ cm. | 22 cm. |
| Hand and forearm (olecranon to tip fifth finger) | 32 cm. | 33 cm. |
| Circumference of wrist (at styloid process) | 16 cm. | 16 cm. |
| Thickness of wrist (at styloid process) | 4 cm. | 4¼ cm. |
| Breadth of wrist (at styloid process) | 5.8 cm. | 6½ cm. |
| Humerus (from acromion to external condyle) | 29½ cm. | 29½ cm. |

There is to be noted a decided shortening of the radius (from a normal of approximately 22 cm.) of 4 cm. on the right, and 3½ cm. on the left, and also a shortening of the ulna (from a normal of approximately 25 cm.) of 4 cm. on the right, and 3 cm. on the left. Motion is the same either active or passive, and is somewhat painful at the extremes. Mobility at the wrist is only slightly affected, and, although the deformity is clinically symmetrical, the right side is affected more than the left. The limit of motion from a horizontal plane is estimated as:

| | Right | Left |
|---|---|---|
| Flexion | 90° | 60° |
| Extension | 30° | 40° |
| Adduction | 30° | 40° |
| Abduction | 25° | 40° |

Pronation and supination are within normal limits.

During a short stay in the hospital for the purpose of observation, a laboratory study of the patient was carried out. Similar studies, partial or complete, have been made by Beder,[10] Brown,[21] Catterina,[40] Fazio,[80] Siegrist,[213] Stetten,[221] Tollas,[227] and Vianna.[231] The results obtained by these investigations indicate that this condition is not manifested by any deviation of the body's chemical or biologic processes. The data acquired in the present instance tend to confirm that impression.

*Laboratory Data.*—Wassermann test negative. Basal metabolism minus four. Uranalysis: Color—yellow; 1,015; alkaline; albumin, sugar, blood and bile, all absent. Microscopic examination essentially negative. Hematology: R.B.C., 5,030,000; Hb., 90 per cent; W.B.C., 8,950. Differential: polymorphonuclears, 65 per cent, lymphocytes, 33 per cent, transitionals, 2 per cent; platelets, 210,000; fragility, 0.45 to 0.27; sedimentation time, 18 Mm. in one hour and 20 minutes.

Blood Chemistry

| | | | |
|---|---|---|---|
| Urea nitrogen | 12.9 mg. | Total cholesterol | 105.3 mg. |
| Sugar | 88.9 mg. | Cholesterol ester | 65 per cent |
| Albumin | 9.3 mg. | Calcium | 10.5 mg. |
| Globulin | 2.2 mg. | Phosphorus | 4.8 mg. |
| Chlorides | 625.0 mg. | Icteric index | 3.8 mg. |

van den Bergh: Direct and indirect—negative.

FIG. 2.—Lateral roentgenogram of the wrist showing the anterior bowing of the radius, and the backward luxation of the head of the ulna.

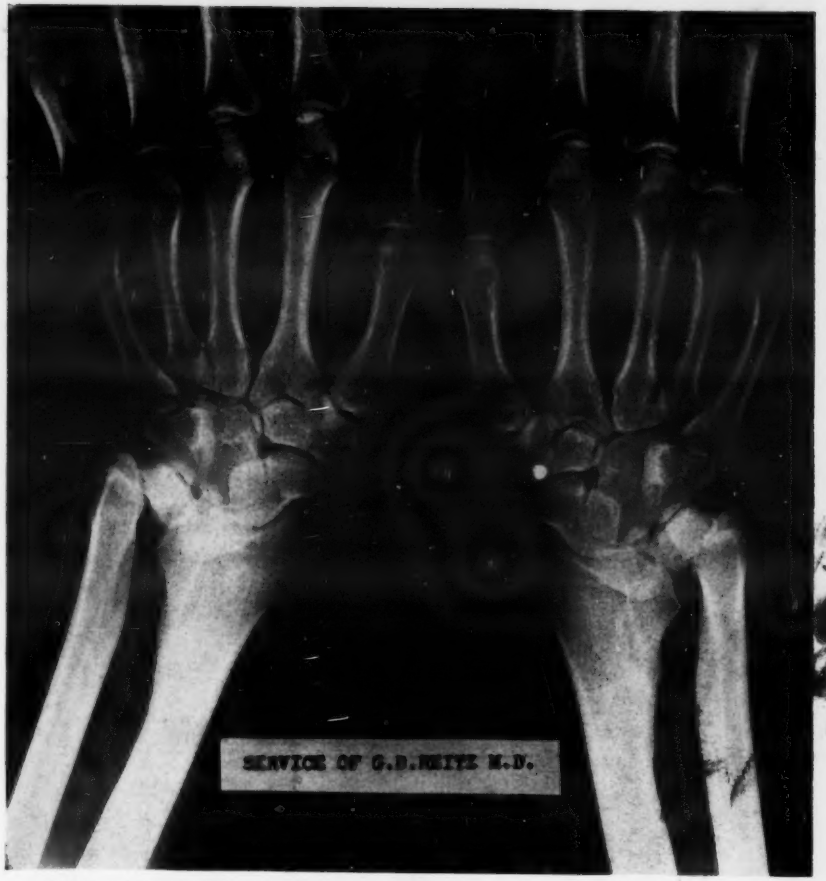


FIG. 3.—Anteroposterior roentgenogram of the wrist and carpus showing the "pyramidalization" of the carpal bones, the absence of the medial half of the epiphyseal line, and the rarefaction of the medial border of the lower end of the radius.

*Roentgenologic Examination.*—Dr. G. H. Kojransky: "There is a striking alteration in the relative lines of the radius and ulna on both sides, the tip of the ulna being on a definitely higher level than the styloid process of the radius. The epiphyseal line of the distal radial epiphysis is visualized on both sides only in its radial half, and instead of running transversely through the whole thickness of the radius, makes a sharp bend, almost at right-angles, in the cephalad direction. The articular surfaces of either radius, as visualized from lateral exposure, are definitely facing forward. The distal ulnar extremity is subluxated in the ulnar-dorsal direction. A small exostosis is noted on the ulnar aspect of the radius opposite the missing half of the epiphyseal line. The roentgenographic appearance is typical of the so-called Madelung's deformity" (Figs. 2, 3, and 4). A roentgenogram of the skull (Fig. 5) revealed: "The character of the digital impressions are very marked, considering the age of the patient, and assume the appearance of the so-called *'lückenschädel.'* There is also evidence of a small amount of calcification in the region of the pineal body. The sella turcica is apparently normal in shape, size and appearance. Roentgenograms of the other long bones showed no abnormalities."

FIG. 4.—Roentgenogram of the entire forearm showing an increase in the normal lateral curvature of the radius, the rarefied area on the internal margin of the lower end of the radius, and the exostosis on its ulnar border.

Because of the increased digital markings of the skull, it was desired to determine if there might be an increase of intracranial pressure. The patient, however, refused to submit to a spinal tap. Examination of the optic fundi did not reveal any papilledema.

In view of the fact that the patient presented no menstrual disorders, or any other evidence of a hormonal dyscrasia, it was not considered necessary to undertake the complicated determinations of the various sex hormones.

The patient was discharged and advised to return at a later date (on cessation of growth) for operative correction of the deformity.

PATHOLOGY.—The data considered in determining the pathology of this condition were obtained by reviewing the 171 cases tabulated herewith, which we have accepted as authentic examples of the deformity. To earlier investigators[89, 115, 135, 158, 192] the opportunity for necropsy diagnosis presented itself, and served to dispel the idea that the deformity was a dislocation of the hand. In 1897, Jagot[114] first appreciated the importance of the roentgenologic examination in determining the diagnosis and pathology of this condition, and this aspect has been progressively developed, as evidenced by the increasing prominence given it in literature.

A review of the published articles reveals that this deformity of the wrist may affect any, or all, of the various structures that go to make up the wrist joint, but it is the *radius*, especially its lower extremity, that is essentially the seat of the primary pathologic phenomenon which is in the nature of an osteochondritic dysplasia, while any abnormalities of the ulna, carpal bones, articular cartilages, ligaments and tendons about the joint, are apparently all of a compensatory nature and secondary to the deformity of the radius.

*The Radius.*—There is a variable widening of interosseous space, due to an exaggeration of the usual, normal lateral curvature of the diaphysis. The most important disturbance is the palmar bowing of the distal end of the radius, usually in the lower third, its greatest degree being situated at the region of the epiphysis or metaphysis, sometimes as if the epiphysis itself had rotated (Fig. 6).

Kirmisson,[122] Stetten,[220, 221] Peckham and Hammond,[166] Gaudier,[96] and Burrows[34] have each reported a case where the usual anterior bowing is

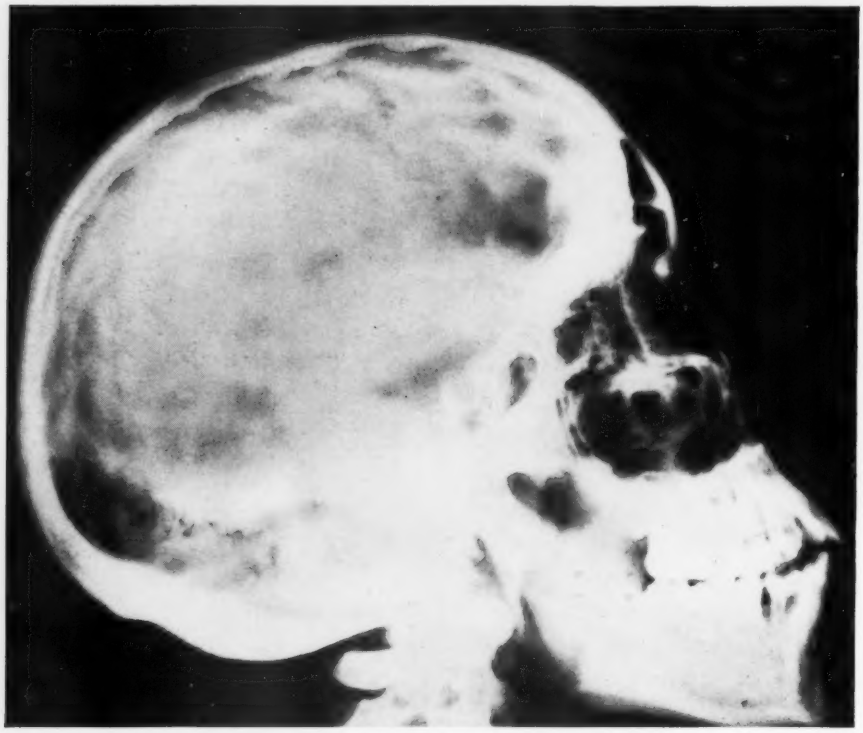

FIG. 5.—Roentgenogram of the skull showing a marked increase in digital impressions, calcification of the pineal body, and a normal sella turcica.

reversed. These constitute the only five authentic cases of posterior bowing or "reverse" type of the deformity (Fig. 7).

The epiphysis has been described as irregular, scalloped and under- or overdeveloped. The epiphyseal line sometimes is found to be broadened, hazy, or entirely absent. If the roentgenograms are examined carefully, it can frequently be appreciated that the epiphyseal line is present only in its lateral half, and its absence in the inner, or ulnar half, is indicative of premature, partial fusion of the shaft and epiphysis. This finding will be noted in the roentgenograms illustrating the present case report.

Another interesting fact, brought out by Rocher[195] and stressed recently by other French authors,[39, 194, 196, 197, 198, 202] is a hemiatrophy of the internal

half of the radial epiphysis. This is indicated on the roentgenogram by a small, rarefied area on the internal, or ulnar side of the radial shaft, immediately above the missing half of the epiphyseal line. This finding was also present in the case herewith reported, and was especially marked on the left side

DIAGRAMMATIC SKETCHES ILLUSTRATING THE ANATOMIC RELATIONS OBTAINING IN MADELUNG'S DEFORMITY IN BOTH THE APPARENT ANTERIOR AND POSTERIOR ("REVERSE") DISLOCATIONS

FIG. 6.—Showing the deformity at the back of the wrist, the bowing of the radius and the apparent anterior dislocation. (A) Section through the radius. (B) Section through the ulna.

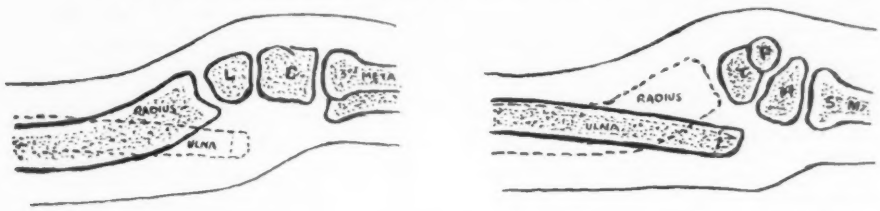


FIG. 7.—Showing the apparent posterior dislocation of a "reverse" Madelung's deformity. (A) Section through the radius. (B) Section through the ulna.

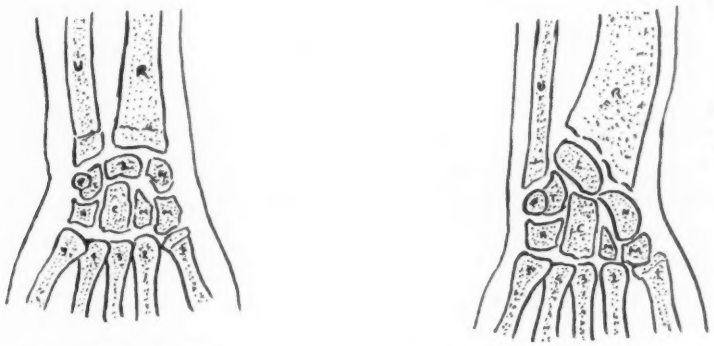

FIG. 8.—(A) Showing the normal arched arrangement of the carpal bones. (B) Showing the "pyramidalization" of the carpal bones in Madelung's deformity, with the os lunatum at the apex.

A third interesting fact, which Stetten[221] describes "as a remarkably frequent finding as a coexisting lesion," is the discovery of an exostosis near the distal epiphyseal line, usually on the ulnar side. These three factors are emphasized, and their significance will be appreciated in the consideration of the pathogenesis.

In addition to any apparent shortening of the total length of the radius due to an anterior or lateral curvature, the radius often suffers an actual

shortening in length, and the patient presents obviously foreshortened forearms.

*The Ulna.*—Although slight bowing and shortening of the ulna has also been occasionally described, this is never more marked than the radius, rarely even as much. More usually the ulna grows forward, unaffected by the pathologic radius, thereby leading to a luxation or subluxation of the inferior radio-ulnar articulation. Most descriptions state: "The ulna is dislocated backward from the radius." Rocher, however, has pointed out that, strictly speaking, it is the radius, and not the ulna, which is dislocated. A hyperostosis of the ulnar head is a common finding.

The bowing and shortening of the radius results in the ulna overriding the carpus dorsally. This gives rise to the most prominent sign of the deformity—swelling at the back of the wrist due to the prominence, or projection, of the ulnar head or styloid. Since the ulna is now at a higher level than the hand and carpus, it gives one the impression of an anterior dislocation. In the "reverse" type of the deformity, the ulna projects below the palmar surface of the wrist and results in the appearance of an apparent posterior dislocation of the hand and carpus. The end of the ulna may be forced back into position in cases of mild luxation, but the dislocation returns on release of pressure. The radial deformity cannot be reduced by manipulation, since the deformity is an integral part of the bone.

*The Radiocarpal Articulation.*—Considering the fact that the radius is deformed in such proximity to the wrist joint, it is inevitable that, in direct proportion to the degree of deformity, the wrist joint will be modified anatomically and functionally. Since the lower end of the radius, which goes to make up the proximal half of the joint, has been bent inward (due to lateral bowing) and downward (or upward in the "reverse" type), the articular surface of the lower end of the radius comes to face downward and somewhat ulnarward, instead of directly forward. Siegrist[212] describes this as "a deviation of the joint surface on two axes," and his article gives a comprehensive discussion of the mechanics of the modified joint.

Since the inferior radial articulation faces downward, instead of directly forward, an anterior displacement of the hand and wrist takes place. There is no true luxation of the joint whatsoever, since the lunate and navicular bones retain their normal relationships to the articular surface of the radius.

Mobility at the joint is modified by the disturbance of mechanical factors. Thus, the anterior bowing of the radius leaves the joint with an apparently hypertrophied superior lip, thereby limiting extension somewhat. The seeming compensatory atrophy of the inferior lip often augments the range of flexion. In the "reverse" type of bowing, the converse is present, and flexion is found to be limited. Depending on the amount of lateral bowing, adduction is variably restricted. The position of the ulna largely decides what range of abduction may take place. Usually the projecting end of the ulna acts as a splint along the lateral border of the carpus, greatly restricting abduction. Occasionally the projection may so disorganize the lateral side of the joint

that the carpal bones are forced into permanent adduction, and render abduction impossible.

Although the pivot of pronation and supination is at the proximal radio-ulnar joint, both of these motions, particularly that of supination, are variably limited because of: (1) The luxation at the inferior radio-ulnar joint. (2) The bowing of the radius. (3) The disparity in size between the radius and ulna.

It is obvious, therefore, that restrictions, limitations and exaggeration of motion about the wrist are directly dependent upon bony impingements. A laxity of the joint membranes and ligaments, which has been a constant finding on dissection, is a negligible factor. Outside of the fact that the flexor tendons are rendered more prominent by being drawn taut over the inferior margin of the articular surface, they do not enter, essentially, into the pathology of this deformity.

*The Hand and Carpal Bones.*—The deviation of the articular surface inward leads to a modification of the usual arched arrangement of the carpal bones, which become wedged in between the deformed radius and the protruding ulna, assuming a triangular appearance, with the os lunatum at the apex (Fig 8). Benneke[13] described this "pyramidalization" of the carpal bones as compensatory to the malarrangement of the component parts of the wrist joint. Slight irregularities of the individual carpal bones have been described.

When the course of the deformity has reached its culmination, the hand and carpal bones come to lie at a lower level than that of the ulna, the hand and forearm assuming a curious and characteristic "bayonet-shaped" deformity (Fig. 1[A]). This has been persistently described as a "silver-fork" (*"gabelhand," "manus furca"*) deformity, but it is obviously a misnomer, as can be seen by viewing the wrist from the side. The exception would be those rare cases of the "reverse" type of the deformity, which is really a "silver-fork" deformity. It was probably Stetten's[220, 221] report describing a case of this type that prompted subsequent authors to misapply that name to the usual deformity.

Of the 171 cases herewith tabulated, it will be noted that the deformities were bilateral in 127 cases, of which 35 were more marked on the right and 21 more marked on the left. Two reports did not state whether a bilateral or unilateral condition existed. Forty-two unilateral cases are reported, of which 18 occurred on the right and 22 on the left side, while in two reports, the side affected was not specified. When both sides are involved, one side usually becomes deformed before the other.

Predisposing Factors.—*Sex Incidence.*—The influence of sex is indisputable—females predominating. The ratio of female cases to male is variably put at 2:1 by Madelung,[135] 31:8 by Abadie,[1] 47:9 by Homuth,[109] 7:1 by Stetten,[221] and 4:1 by Salisachs.[203] Analysis of the 171 cases considered in Table I shows a ratio of 137:33 (one not specified), or an approximate ratio of 4:1.

*Age Distribution.*—This condition is, essentially, an affection of adolescence,

occurring in the secondary growth period, and has its inception, with but few exceptions, between the ages of ten and 14.

*Heredity.*—Definite instances of a hereditary influence are to be noted in nearly one-third of the reported cases. Guépin[104] reported a case of a girl whose mother presented an even more marked deformity; the father also had large wrists; the sisters and brothers of the mother showed a similar lesion; all nine sisters and brothers of the patient and the children of a sister had had prominent ulnae from childhood. In Jagot's case,[114] the father, uncle, and grandfather had thickened wrists, but did not have any functional disturbance. Other instances of the deformity in three generations are to be found in the reports of Roget[200]—mother and grandfather; Volkmann[234]—mother and grandmother; Siegrist[212]—sisters, mother and grandmother; and Reich.[188] Cases occurring in siblings have been described by Ardouin[7]—brothers; Solberg[214]—brothers, whose parents were cousins; Sauer[204]—brother and sister; Pedrazzi[167]—mother and sister; Vidal[232]—mother and sister; and Wéry[236]—sister. Not infrequently the deformity is present in mother and daughter, as cited in the reports of Dekeyser,[59] Gangolphe,[92] Malfuson,[137] Levyn,[132] and in Stetten's case,[221] where there was a history of deformed wrists in the mother. Other instances of a hereditary nature are found in the reports of Estor,[77] Brandes,[26] Salisachs,[203] Gaugele,[98] Magnus,[136] and Massabuau.[143] Nove-Josserand, in the discussion of Gangolphe's[92] paper, suggests that a latent heredity, becoming active when the hands are first actively used, may be the etiologic factor of the deformity.

*Occupation.*—This factor was once stressed by the early French writers, but it is illogical to suppose that any condition occurring usually in young girls who are either too young to work or whose work is of a comparatively light nature can claim occupation as an etiologic factor.

*Environment.*—The high percentage of cases reported from the poorer classes can be easily explained by the fact that it is this class of patients that supply the clinical material upon which most of our medical literature is based. It was the opinion of the early writers that living conditions provided an etiologic factor to the development of this condition comparable, possibly, to the occurrence of rickets, tuberculosis, *etc.*

PATHOGENESIS.—The multiplicity of nomenclature designating this condition reflects the variable pathologic processes propounded, and it has been remarked that there are nearly as many etiologic theories as there are authors. As examples, the following are instructive: "Manus valga" (Madelung, Siegrist and Sauer); "carpus curvus" (Delbet); "radius curvus" (Destot, Gangolphe); "congenital dislocation of the wrist" (Pooley, Estor); "late rickets of the wrist" (Duplay, Salisachs); "progressive subluxation of the wrist" (Kirmisson); "progressive idiopathic curvature of the radius" (Stetten); "adolescent club-hand" (Mauclaire); "carpo-cyfose" (Robinson et Jacoulet); "cubitolisthesis" (Palazzi); "manus furca" or "gabelhand" (Springer); "radius brevior" (Masmonteil); and "dyschondroplasia of the inferior radial epiphysis" (Rocher).

ETIOLOGIC THEORIES.—The following theories have been advanced explaining he etiology of Madelung's deformity, some of which are no longer tenable, but are included for the sake of historic completeness:

(1) *Trauma.*—This factor was advanced by the old French school (Bégin,[11] Dupuytren[73]). Along with the predisposing factor of occupation, trauma has been dropped from serious consideration for much the same reason—its preponderant occurrence in young girls, who are certainly less exposed to trauma than the general population.

(2) *Muscular.*—Madelung[135] himself, gave credence to this theory when he stated: "The main factor in its causation is the more powerful action of the flexors of the forearm, due to overexertion. Continued hyperflexion stretches the extensor tendons and the posterior ligaments over the dorsum of the radial epiphysis, exerting a forward force and producing a volar bowing." Busch,[35] who championed this theory, attempted to correct the deformity by tenotomy of the flexor tendons, but the procedure proved to be ineffectual. This theory could hardly explain those cases of posterior bowing, nor does it take into account the anatomic position of these muscles and the insertions of their tendons, which would lead one to expect deformity to take place in the fingers and hand, rather than at the wrist.

(3) *Nervous.*—This theory, advanced in a hypothetic manner by Félix,[81] is altogether untenable, and is merely mentioned in passing.

(4) *Osseous Dystrophies.*—Landivar[125] compares the deformity to the group of conditions such as Paget's or von Recklinghausen's disease, *etc.*, but these conditions are each of their own clinical entity, and occasionally, if they should present a "symptomatic" form of Madelung's deformity, as in Bjorkroth's[23] or Rocher's[193] case, they may be easily differentiated roentgenologically.

(5) *Inflammatory.*—Considering the fact that osteomyelitis, as well as Madelung's deformity, also frequently occurs during the secondary growth period, and that Rosenow has succeeded in demonstrating organisms specific for various parts of the body, it is surprising that this theory has had such fleeting and ineffectual backing. However, this is due to a lack of any positive evidence in its favor.

(6) *Endocrine.*—It is inevitable that the preponderance of female cases would suggest an endocrine basis for the condition. Earl,[74] Cserey-Pechany,[51] and Beder[10] have reported cases of the deformity which showed mild or severe ovarian hypofunction. Cserey-Pechany gave his two patients ovarian and glandular hormone therapy for a period of seven months. He not only claimed to have checked the course of the disease, but attempted to demonstrate roentgenologically an actual regression of the deformity. However, the course of the disease may cease at any time, even without medication, just as spontaneously and as insidiously as it began. As for the roentgenographic evidence, one must realize that, unless serial exposures are made at exactly the same angle, marked differences in the appearance of the curvature may result.

(7) *Rickets.*—This theory has been regarded as the most frequently pro-

pounded and ardently defended of all. A canvass of the literature reveals a comfortable majority as proponents of the etiologic factor of tardy, or late, rickets. Salisachs[203] is a contemporary exponent of this theory, and a large part of his article is devoted to an attempt to prove its applicability. In rebuttal, we would like to point out that rickets is a constitutional disease, and shows other manifestations besides bony pathology, which are never seen in Madelung's deformity, nor have there been more than one or two cases in all those reported of the condition where other bony manifestations, such as craniotabes, Harrison's groove, rachitic-rosary, *etc.*, have been demonstrated. The occasional coexistence of scoliosis can be explained on the ground of posture, nor is it unusual to find scoliosis coexisting with practically any study made of dispensary patients. The actual existence of rachitis tarda as a clinical entity has been denied by several authors (Holt[108]), but even if one were to presume its existence, it would be difficult to explain how it would localize itself to one or both radii, to the exclusion of the rest of the skeletal system, since even those authorities who do believe in the existence of late rickets (Clutton,[45] Roose[201]) insist that general epiphyseal lesions are essential for a diagnosis.

(8) *Dyschondroplasia of the Distal Radial Epiphysis.*—Even Madelung, in his original presentation, presupposed "a primary weakness of the bone" in the etiology of the deformity, while Redard[186] expounded the idea that the disease was due to a disturbance of growth of the epiphyseal cartilage. Delbet[60] also concluded that it was due to an irregular development of the epiphyseal cartilage, related to cartilagenous exostosis, analogous to genu varum, but, like Redard, he imputed it to late rickets. Stetten[221] states that: "Closer to the correct solution comes the suggestion that the deformity is a disease of the epiphyseal cartilage," and rejects entirely the factor of late rickets.

The mechanism of dyschondroplasia is suggested by the rarity of cases; the remarkable uniformity of the age at the onset; the usual bilateral occurrence; the absence of the usual stigmata of rickets or a history of local trauma; its onset during the secondary growth period; and the frequent association of the deformity with other anomalies. Kun[124] and Ingber[112] report the coexistence of an asymptomatic sacralization of the fifth lumbar metamere; Kajon,[119] an homolateral cervical rib; Beder,[10] Brown,[31] Stokes,[223] Mathieu,[145] von Bergmann,[17] and Chierici,[41] bowing of the tibiae; Rocher,[197] absence of the caput humerus; Kun[124] and Vianna,[231] spina bifida occulta; von Bergmann[17] and Melchior,[151] brachymetacarpals; Solberg,[214] Franke,[87] and Fazio,[80] stunted growth or dwarfism; Gadrat,[90] Curtillet[52] and Dimitriu,[69] multiple exostosis. Possibly if all the cases reported were to have had the entire skeleton examined roentgenologically, numerous other osteochondritic anomalies would have been discovered.

Bessel-Hagen[19] has attempted to demonstrate an arrest in the development of any bone which gives rise to exostosis formation. Stetten[221] remarks on the frequency of finding an exostosis near the distal epiphyseal line as a coexisting lesion in Madelung's deformity. The case herewith reported

presents this lesion, which may be noted on the ulnar side of the radius, at the level of the missing epiphyseal line (Fig. 3). According to the observations of Bessel-Hagen, this would lead to an arrest of development on that side, which is apparently what happens. The lateral side of the radius continues to grow at the epiphysis, sweeping the outer margin of the radius along an arc, thereby giving rise to the lateral bowing of the radius. This lateral curvature was stressed particularly by Duplay,[72] who ascribed it to rickets.

If, as we are inclined to believe, the formation of the exostosis is secondary to, or part of, a premature fusion of part of the epiphysis with the diaphysis, there will result a local cessation of growth. Growing ends of bone are known to receive their blood supply from four sources: (1) The large nutrient artery which supplies the diaphysis; (2) a comparatively small epiphyseal artery; (3) the periosteal vessels, which supply the cortex; and (4) the juxta-epiphyseal vessels, as described by Lexer, which supply the growing metaphysis.

Thus, with premature fusion of part of the epiphysis, this last source of nutrition is cut off, and its loss is indicated by a comparatively rarefied area of bone. This rarefied area, to be found on the ulnar half of the radius, just above the epiphyseal line, is well shown in Figure 3, and has been repeatedly stressed by Rocher[194, 195, 196, 197, 198] and by Canton.[39]

The hypothesis of local and partial fusion of only a section of the epiphysis can be deduced from the frequent finding that the epiphyseal line is indistinct, or even partly missing, as is shown in Figure 2. If we apply the same train of thought to early fusion of the volar half of the inferior radial epiphysis, we can see that the anterior bowing of the radius may be the result of local "achondroplasia," and that the mechanism of anterior bowing is dependent upon cessation of growth in the volar half of the epiphysis. This is far more logical than the usual hypothesis, promulgated by Gangolphe,[92] that the pressure of flexor action leads to an atrophy of the anterior half of the epiphyseal cartilage, with a compensatory hypertrophy of the posterior half.

Pels-Leusden[168] also presented the hypothesis that a disease of the intermediary cartilage caused premature ossification of the ulnar and volar side, with a change in the direction of longitudinal growth. Gickler[102] proposes to explain this premature partial fusion on the ground of a hemorrhage into the metaphysis as a result of disease, weight-bearing (as in crawling about in infancy), slight trauma, or even rickets. Hemorrhage would tend to cut off the local blood supply and lead to early fusion. Redard[186] had previously observed that continuous irritation, such as might be received from performing some particular kind of work (washing, wringing, *etc.*) produced a functional hyperactivity of certain portions of the cartilage.

Fick and Pahil[84] point out that the dyschondroplasia itself is the basic factor, and any curvature produced is merely accidental, depending upon the portion of the epiphysis which undergoes dyschondroplasia. Cases have been reported by Gickler,[102] and Dee[57] where no bowing of the radius existed, but where

dyschondroplasia was present. Any early fusion due to dyschondroplasia will give rise to an actual shortening in the length of the radius.

We mention, in passing, that the mechanism of bowing in the rarer "reverse" type of the deformity is the premature fusion of the dorsal half of the radial epiphysis leading to posterior bowing. Stetten[221] considers the site of bending of the radius dependent upon the age at which the disease begins, the nearer the lower end of the shaft, the later the affection. Where the entire radius is curved, the process must have begun during intra-uterine life, although a visible deformity may not have been noted until the deformity reached its height. The degree of curvature would depend upon the activity of the pathologic process.

DIFFERENTIAL DIAGNOSIS.—From the foregoing description, it would seem to be quite easy to diagnose this condition, but the large number of similar and secondary deformities which have been improperly introduced into the various bibliographies belies that ease. It is quite possible that fracture, dislocation, arthritis deformans, osseous dystrophies, tumor formation, *etc.*, might produce the bayonet-shaped deformity itself, but a careful history should elicit the spontaneous, idiopathic onset of the disease during the secondary growth period, its progressive nature giving an early clue to the diagnosis. A complete physical examination will reveal an absence of rickets and other general, or local, disturbances. A roentgenologic examination of the lesion would seem necessary to rule out a "symptomatic" form, and would definitely confirm the diagnosis beyond a doubt.

Those authors who have reviewed the literature extensively are almost unanimous in expressing a plea to separate the genuine Madelung's deformity from a "pseudo,"[41] "spurious,"[153] "symptomatic,"[23] "simulating,"[70] "atypical"[228] or "similar conditions,"[34] and other unrelated types of deformity of the wrist which have masqueraded into the literature of the true variety. Several authors, in deploring this hegira from specificity, have attempted to correct it by adding still more to an already overburdened nomenclature. Zeitlin[239] suggests "Madelung's disease" (morbus Madelung), but Madelung has already given his name to a disease characterized by a diffuse symmetrical limpomatosis, or deposit of fatty tissue, on the upper part of the back, shoulders and neck. Burrows[34] suggests "Madelung's syndrome," but a deformity is not, strictly speaking, a "syndrome," and it has all the faults of eponymous nomenclature.

In condemnation of the pernicious custom of applying a person's name to a pathologic condition or operation, often undeservedly, we would suggest that Madelung's name be dropped. Sir Jonathan Hutchinson[110] has remarked: "So in pathology we have to contend against the tendency to substitute a name, or a definition, for the perception of an essential nature. As it is easier for us to worship a name, or even a book, rather than to conceive of a nonmaterial power, so we more readily become accustomed to content ourselves with some euphonious name for a disease, rather than to acquire the habit of constantly trying to realize its nature and its relation to possible causes."

The Standard Classified Nomenclature of Disease[219] has a system of

nomenclature based on both a topographic and an etiologic factor. Following their method of classification, Madelung's deformity would be indexed: 23132-077 (that is: distal radial epiphysis—dyschondroplasia of) and we, therefore, suggest calling the deformity "Dyschondroplasia of the Distal Radial Epiphysis."

Several writers, with whom we are inclined to agree, place this deformity among the better known, but equally cryptogenic, group of conditions which includes Perthe-Calvé-Legg's, Osgood-Schlater's, Köhler's, and Keinboch's diseases. We suggest that these diseases be classified under the head of dyschondroplasias, and that they be renamed, as, for example, dyschondroplasia of the caput femoris instead of Perthe's disease, or scaphoid dyschondroplasia instead of Köhler's disease, *etc.*

Berg[16] and Ewald,[79] among others, challenge the right of Madelung's deformity to be considered a clinical entity, and advance the opinion that the deformity is merely the sign of some underlying pathology—such as rickets, lues, osteitis deformans, *etc.*

Classifications of Madelung's deformity have already been advanced by Abadie,[1] Barthés,[9] Depage,[64] Gasne[95] and Stetten.[221] It would seem advantageous, considering the present state of the literature, to formulate a classification that will include all the various types of the deformity, including the secondary static types. The following classification is, therefore, presented:

A. Presenting Radial Deformity:
   (1) With anterior bowing of the radius:
      (a) Radial dyschondroplasia (genuine Madelung's deformity).
      (b) Secondary static deformity: Traumatic;[70, 237] luetic;[76] inflammatory;[64, 21] tuberculous;[38] osteitis;[233, 193] rickets,[91] *etc.*
   (2) With posterior bowing of the radius:
      (a) Radial dyschondroplasia ("reverse" Madelung's deformity).
      (b) Secondary static deformity.[150]
   (3) Without bowing:
      (a) Radial dyschondroplasia.[57, 102]
      (b) Secondary static deformity.
B. Presenting Ulnar Deformity:
      (a) Ulnar dyschondroplasia.[238]
      (b) Secondary static deformity.[162]

Prognosis and Treatment.—Pain may be present as long as the pathology progresses, but ceases when growth is arrested at the wrist, usually before the age of 25. The patient may then be educated in the better use of the deformed wrist.

Treatment, at least up until the cessation of growth, should be palliative, and consists in resting the part. This may be effected by splints, braces, or a plaster encasement, but these measures will not correct the deformity, nor will they prove effective in halting the progress of the disease. Tenotomy of

the flexor tendons of the forearm is unjustifiable, and the mere removal of the distal end of the ulna is also condemned.

Osteotomy of the radius for the correction of the deformity has proven so effective, in competent hands, that it may be recommended routinely, just as one would in a case of badly bowed legs, malunited fracture, *etc.* In 1885, Duplay[71] tried a transverse linear osteotomy; in 1904, an oblique type was advocated by Poulsen.[178] Although these operations may suffice for mild forms of the deformity, most cases may necessitate a cuneiform osteotomy. The curve in the lower end of the radius is corrected, and a plaster encasement is applied, with the hand put up in extension and abduction. Springer,[218] advises that the hand be forced, if necessary, into supination (depronation). In order to hold the fragments more securely, Taylor[225] inserted a metal plate.

When the ulna is markedly longer than the radius after the osteotomy, its projecting end must be resected in order to obtain a perfect functional result. However, since an osteotomy is liable to shorten an already stunted radius, Burrows[34] has devised an ingenius procedure wherein the resected head of the ulna is shaped and inserted as a bone graft, or peg, between the fragments of a linear osteotomy, thereby tending to conserve, or at times elongate, the length of the radius. The postoperative care is the same as for any osteotomy.

## SUMMARY

(1) A typical case of Madelung's deformity is presented; the twenty-first to be reported from this country.

(2) The literature on this subject has been reviewed, and is found to be inaccurate, incomplete and is replete with reports of cases that are not genuine. An attempt has been made to correct these errors and to properly correlate the history of this condition.

(3) A tabulation of 171 authentic cases has been compiled.

(4) We advocate the substitution of the term "Dyschondroplasia of the Distal Radial Epiphysis" in place of "Madelung's deformity," since the latter term is not specific.

(5) A classification of the various types of the deformity has been formulated.

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[81] Félix, Joseph: Étude sur la subluxation spontanée du poignet en avant. Thèse de Lyon, 246, 1884. (2 cases.)

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[87] Franke: Zur Anatomie der Madelung'schen Deformität der Hand. Deutsche Zeitschrift für Chirurgie, **92,** 156–180, 1908. (1 case, with dissection. This is the same case as that by Müller.[155])

[88] Frölich, M.: Radius Curvus ou Maladie de Madelung. Revue Médicale de l'est, **1,** 586–587, 1922. (Discussion on Mathieu's[144, 145] case.)

[89] Frölich, M.: Discussion on Finzi's[85] paper. Seventeenth International Congress of Medicine, London, 1913, **7**, Part 2, Orthopedics, pp. 344. (Case described is too indefinite for inclusion and is probably that of Mathieu and Joseph,[144, 145] q.v., upon which he operated.) Also see La Presse Médicale, **21**, 592, 1913. (Case 1, as above; Case 2 arthritis; Case 3, osteomyelitis.)

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[92] Gangolphe: Déformation singulière du poignet inexactement denommée subluxation spontané. Bulletin de la Société de Chirurgie de Lyon, **2**, 117–123, 126, 135, 1899. (2 cases.)

[93] Gangolphe: Malformation congénitale du poignet. Lyon Médicale, **90**, 451, 1899. (Same cases as above.[92])

[94] Garrido-Lestache, J.: Un caso de enfermedad de Madelung. La Pediastria Española, **14**, 138–143, 1925. (1 case.)

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[97] Gaugele, Karl: Madelung'schen Handgelenks Deformität. Archiv. für Klinische Chirurgie, **88**, 1058–1075, 1909. (Cases 1 and 2, no bowing of radius Case 3, ref. 98.)

[98] Gaugele, Karl: Gibt es eine genuine Madelung'sche Handgelenks Deformität? Zeitschrift für Orthop. Chirurgie, **24**, 462–479, 1909. (4 cases; 3 are accepted; the fourth was rachitic.)

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[100] Gery, de Chastenet et Colombier: Deux cas de Radius Curvus. Bull. et Mém. de la Société d'Anatomie de Paris, **17**, 370–376, 1920. (2 cases, with dissection of Case 1.)

[101] Gevaert, G.: Un cas de subluxation du poignet de Madelung. Revue d'Orthopédie, 2nd series, **3**, 335–342, 1902. (1 case.)

[102] Gickler, H.: Wachstumsstörung der Radiusepiphyse und Madelung'sche Deformität. Archiv. für Orthop. und Unfall Chirurgie, **33**, 312–318, 1933. (4 cases of dyschondroplasia of the inferior radial epiphysis, but with no bowing of radius.)

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[104] Guépin, A.: Laxité congénitale de l'articulation radio-cubital inférieure et subluxation consecutive de la tête du cubitus en arrière. Comptes rendus hebdonnadaires des séances et mémoires de la Soc. de Biologie, **44**, 627–631, 1892. (2 cases with a history of similar deformity in 14 members of the family.)

[105] Guéry, A.: Un cas de luxation progressive du poignet (Subluxation spontanée de Madelung). Revue d'Orthopédie, **9** 277–282, 1898. (1 case.)

[106] Guye: Observation d'un cas de maladie de Dupuytren-Madelung bilaterale. Revue Médicale de la Suisse Romane, **39**, 191–192, April, 1919. (1 case.)

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[111] Ianni, Raffaele: Radius Curvus; Deformità di Madelung-Duplay. Annali Italiana di Chirurgia, **3**, 40–61, 1924. (1 case.)

[112] Ingber, E.: Bilateral Madelung Deformity and True Asymptomatic Sacralization of the fifth Lumbar Metamere; Roentgen Study of a Case. Quaderni Radiologia, **5**, 251–257, 1934. (1 case; not obtainable.)

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[114] Jagot, C.: Sur une vice héréditaire de conformation des deux poignets. Archives Méd. d'Angiers, **1**, 159–170, 1897. (1 case.)

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[117] Jósa, László: A csuklo Madelung-féle deformitasanak egy esete. Orvosi Hetilap, **70**, 1321–1324, 1926. (1 case.)

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[119] Kajon, Cesar: Madelung'sche Deformität konbiniert mit Halsrippen. Wien. med. Wochenschrift, **84**, 460–462, April 21, 1934. (1 case with cervical ribs.)

[120] Kieffer, Charles F.: Congenital Dislocation of Both Ulnae at the Wrists. ANNALS OF SURGERY, **38**, 119, 1903. (1 case.)

[121] Kassowitz, M.: Die Ursache der Gelenkschlaffheit der Rhachitis. Centralblatt für Chirurgie, **9**, 385–390, 1882. (This much quoted article presents no cases nor mentions Madelung's deformity.)

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[126] Laurence, Joseph: La Maladie de Dupuytren-Madelung. Revue Générale de Clinique et de Thérapeutique, et Journal des Practiciens, **37**, 75, 1923. (Discussion.)

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[132] Levyn, L.: Madelung's Deformity; A Report of Two Cases, Constituting the Fifth and Sixth American Cases. Radiology, **3**, 145–149, August, 1924. (2 cases.)

[133] Lladó, Antonio Cortés, y Gallardo, Louis Salvador: Estudio de la anatomia y patogenia de un caso de deformidad de Madelung. Revista Médica de Barcelona, **4**, 251–274, September, 1925. (1 case.)

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[135] Madelung: Die spontane Subluxation der Hand nach vorne. Verhandlungen der Deutschen Gesellschaft für Klinische Chirurgie, **23**, 395–412, 1879. (5 cases.)

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[137] Malfuson, Daniel: Déformation du poignet d'origine probablement rachitique. Thèse de Paris, July 28, 1894. (2 cases.)

[138] Malgaigne, J. F.: Traité des fractures et des Luxations. Paris, **2**, 711–712, 1855. (1 case.)

[139] Marsan, Felix: Sur un noveau cas de maladie de Madelung. Archives Générales de Chirurgie, **2**, 472–482, November, 1908. (1 case.)

[140] Marsan, Felix: La Maladie de Madelung (Radius Curvus). Gazette des Hôpitaux, **81**, 1671–1679, 1909. (No new reports, but an excellent discussion and bibliography up to 1909.)

[141] Masmonteil, Fernand: À propos de la pathogénie de la maladie de Madelung (Radius Brevior). Gazette des Hôpitaux, **93**, 101–103, 1920. (Traumatic deformity.)

[142] Masmonteil, Fernand: Toujours à propos de la pathogénie de la maladie de Madelung. Lyon Chirurgical, **18**, 351–355, 1921. (1 case.)

[143] Massabuau, Soulas et Nichet: La Maladie de Madelung. Archives de la Soc. Méd. et Biol. de Montpellier, **15**, 191–195, May, 1934. (1 case.)

[144] Mathieu, Ch., et Joseph, V.: Radius Curvus Bilateral. Société Méd. de Nancy, May 24, 1922; Revue Médicale de l'est, **1**, 586, 1922. (Same case ref. 145.)

[145] Mathieu, Ch., et Joseph, V.: À propos d'un cas de Maladie de Dupuytren-Madelung bilaterale. Revue Médicale de l'est, **1**, 691–701, 1922. (2 cases.)

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[147] Mauclaire: Trois observations de subluxation progressive du poignet. Comptes rendus de la Soc. de Chir. de Paris, February 9, 1916; Bulletins et Mémoires de la Société de Chirurgie de Paris, **42**, 344, 1916. (3 cases.)

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[152] Melchior, Edouard: Die Madelung'sche Deformität des Handgelenks. Ergebnisse der Chirurgie und Orthopädie, **6**, 649–680, 1913. (Discussion.)

[153] Merlini, A.: La deformità di Madelung. La Chirurgia degli Organi di Movimento, **9**, 245–268, March, 1925. (Case 1—Madelung; Case 2—traumatic.)

[154] Moore, B. H.: Radius Curvus; Madelung's Wrist. Journal of Bone and Joint Surgery, **6**, 568–574, August, 1924. (2 cases.)

[155] Müller, W.: Madelung'scher Deformität des Handgelenkes. Zentralblatt für Chirurgie, **34**, 1333–1334, 1907. (Same case as Franke,[87] *q.v.*)

[156] Natvig, Reinhardt: Madelung's Haanddeformitet. Tidsskrift for dem norske Laegeforening, **25**, 535–555, 1905. (1 case.)

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[158] Nélaton, Ch.: Luxations Radio-carpiens. Traité de Chirurgie, de Duplay et Reclus, **3**, 121–125, 1897. (1 case, described on pages 124–125, is a dissected specimen.)

[159] Nové-Josserand, G.: La maladie de Dupuytren-Madelung. Seventeenth International Congress of Medicine, London, 1913, **7**, Part 1, Orthopedics, 206–208. (Brief discussion; no case report.)

[160] Ollier, L.: Traité expérimental et clinique de la régénération des os et de la production artificielle du tissu osseux. Paris, 1877, **1**, 402. (Basis for Dalbera's[55] work on Ollier's Law.)

[161] Ollier, L.: Traité des Resections, **1**, 408, 1885 (Curved radius due to resection of cartilage); **2**, 441, 1889. (Deformity secondary to osteitis of the lower end of the radius.)

[162] Painter, Chas. F.: Congenital Dislocation of the Carpus. Bryant and Buck's American Practice of Surgery, **4**, 742–746, 1908. (Description meager; apparently an ulnar deformity.)

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[166] Peckham, Frank E., and Hammond, Roland: Madelung's Deformity. Boston Medical and Surgical Journal, **160**, 447–448, April 8, 1909. (1 case; Case 3 acceptable; Case 4 too vague.)

[167] Pedrazzi, Carlo: Deformità di Madelung famigliare. La Radiologia Medica, **14**, 125–132, February, 1927. (2 cases.)

[168] Pels-Leusden: Madelung'sche Deformität der Hand. Freie Vereinigung der Chirurgen Berlins. Zentralblatt für Chirurgie, **34**, 190, 1907. (1 case.)

[169] Pels-Leusden: Über die Madelung'sche Deformität der Hand. Deutsche Med. Wochen., **33**, 372–374, 1907. (1 case.)

[170] Perrin: Exostoses osteogeniques multiples accompanées d'arrets de développement et de deformations du squellete. Revue d'Orthopédie, **5**, 53–82, 1914. (Deformities due to exostoses.)

[171] Pilatte, Réné: Contribution à l'étude du radius curvus. Thèse de Paris, 134, 1919. (3 cases.)

[172] Pilatte, Réné: Sur la pathogénie du radius curvus. Revue d'Orthopédie, **8**, 223–224, 1921. (Discussion.)

[173] Piollet: Un cas de radius curvus. Bulletin de la Société de Chirurgie de Lyon, **9**, 274–277, July 5, 1906. (1 case.) Lyon Med., **107**, 799, 1906. (Same case.)

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[175] Poncet, Antonin, et Leriche, Réné: La Maladie de Madelung. Ses modalities, sa pathogénie. Gazette des Hôpitaux, **82**, 187–191, February 9, 1909. (Discussion of deformity; cases as reported above.[174])

[176] Pooley, J. H.: Congenital Dislocation of the Wrists. The American Practitioner of Medicine, **21**, 216–220, 1880. (1 case.)

[177] Poucel: Carpus Curvus. Séance de la Société de Chirurgie de Marseille, November, 1929. La Presse Médicale, **38**, 24, 1929. (Description too meager for acceptance.)

[178] Poulsen, Kr.: Über die Madelung'sche Deformität der Hand. Archiv für Klinische Chirurgie, **75**, 506–532, 1905. (2 cases.)

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[181] Putti: Sur la malformation de Madelung. Revue d'Orthopédie, **10**, 207–220, 1909. (1 case, museum specimen.)

[182] Quadrone, Carlo: Contribution à l'étude de la Maladie de Madelung (Subluxation spontanée de poignet). Nouvelle Iconographie de la Salpêtrière Paris, **24**, 71, 1911. (1 case.)

[183] Quarenghi, F. M.: Contributo allo studio della cosi delta Malattia di Madelung. Pensiero Med., Milano, **13**, 57, 1918. (Not obtainable.)

[184] Raconski, F. E.: Madelung'sche Deformität; Cas. Soplek. Cesk., **14**, 125, July 15, 1927. (Apparently a traumatic case. Not obtainable.)

[185] Ramos, Alvaro: A proposito de dois casos legitimos da deformação de Madelung. Archives Brasileiros de Medicina, **1**, 1, 1911. (Case 1, Madelung; Case 2, rickets.)

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[190] Robert, César Alphonse: Des vices congénitaux de conformation des articulations. Thèse au concours, 1851. (Not obtainable.)

[191] Robinson: De la carpo-cyphose. Gazette des Hôpitaux, **81**, 1781, 1908. (No new case report.)

[192] Robinson, R., et Jacoulet, F.: La luxation congénitale de l'extrémité inférieure du cubitus. Archives Générales de Chirurgie, **3**, 1–30, 1909. (2 cases; Case 2 dissected.)

[193] Rocher, H. L.: Radius curvus symptomatique d'une ostéité fibreuse de la moitie inférieure du radius droit. Gazette Médicale de France, **10**, 437–439, May 15, 1935. (This is not a true Madelung's deformity, but secondary to osteitis.)

[194] Rocher, H. L.: A propos de notre septième observation de Maladie de Madelung; Dyschondroplasie radio-cubitale inférieure par hémiatrophie epiphysaire radiale interne. Journal de Médicine de Bordeaux, **114**, 513–518, April 17, 1937. (1 case.)

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[197] Rocher, H. L., et Rocher, Christian: La Maladie de Madelung; Dyschondroplasie Radio-cubitale Inférieure Conditionnée Essentiellement par l'Hémiatrophie Epiphysaire Radiale. Chirurgia degli Organi di Movimento, **20**, 20–30, 1934. (1 case; Case 2 described previously.)

[198] Rocher, H. L., et Roudil, G.: Dysmorphose congénitale bilaterale des poignets par hémiatrophie epiphysaire radiale. La Presse Médicale, **38**, 1089–1090, August 13, 1930. (1 case.)

[199] Roederer, C.: Un cas de maladie de Madelung larvée. Bull. et Mém. de la Soc. de Chir. de Paris, **20**, 235–237, March 16, 1928. (1 case; Case 2 too brief for acceptance.)

[200] Roget, Eugene: Étude sur le radius curvus. Thèse de Lyon, 121, April 26, 1899. (1 case.)

[201] Roos, E.: Über späte Rachitis. Zeitschrift für Klinische Medizin, **48**, 120–144, 1903.

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[203] Salisachs, Louis Gubern: Contribuçion al estudio de la deformidad de Madelung.

Revista Médica de Barcelona, **20**, 105–137, August, 1933. (Case 1, fracture; Case 2, rickets; Cases 3, 4 and 5 acceptable.)

[204] Sauer, F.: Die Madelung'sche Deformität des Handgelenkes. Beiträge zur Klin. Chir., **48**, 179–203, 1906. (3 cases.)

[205] Savariaud: Maladie de Madelung-Duplay ou subluxation progressive et spontanée du poignet par radius curvus chez une jeune fille attiente d'ostéité spécifique. La Presse Médicale, **21**, 592, 1913. (Case not accepted due to too meager description.)

[206] Schade, A.: Kasuistischen Beiträge zu den Luxationen im Handgelenke auf Grund pathologischer Zustände. Inaugural Thesis at Kiel, 1906. (1 case.)

[207] Schnek, Fritz: Die operative Besserung der echten und der sogenannten symptomatischen Madelung'schen Deformität. Zeitschrift für Orthopädische Chirurgie, **1**, 320–329, October 14, 1928. (Case 1, Madelung's deformity; Case 2, osteomyelitis; Case 3, Madelung's deformity.)

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[209] Schulze, H.: Ein Fall von spontaner subluxation der Hand nach unten (Dupuytren-Madelung'scher Subluxation). Münchner Medizinische Wochenschrift, **52**, 1441–1443, 1905. (1 case.)

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# INTRACAPSULAR FRACTURES OF THE NECK OF THE FEMUR*

## A STUDY OF ONE HUNDRED NINETY CASES

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EVALUATION of any method of treatment of intracapsular fractures of the femoral neck is exceedingly difficult because of the varied and contradictory reports of end-results found in the literature.

For the past 20 years the Whitman[1] abduction treatment has been the most popular and widely employed method for the treatment of fractures of the hip. Although other methods, including open operation, have been used in a few clinics, it was not until 1931, when Smith-Petersen[2] presented his work on internal fixation, that operative procedures became widely utilized. This widespread acceptance of a new surgical approach to the problem would indicate that the closed methods previously used had proved unsatisfactory.

In 1930, a commission appointed by the American Orthopedic Association[3] investigated the end-results of the Whitman method of treatment of intracapsular fractures, and in a careful study of 210 cases, throughout the country, found that osseous union was obtained in 53.8 per cent of the cases. In 1934, McAusland[4] reported a series of 60 cases treated by the Whitman method in which 58.3 per cent obtained bony union. In the same year Henderson[5] reported that 66.6 per cent of a series of 36 cases treated by the Whitman method resulted in bony union. Howard and Christophe,[6] in 1934, reported 70 per cent bony union in 43 cases treated by the Whitman method. As late as 1936, Whitman[7] stated that the lowest percentage of bony union reported in the series of cases treated by his method was 58 and the highest 90. In the same year Mensor and Dewey[8] reported 50 cases admitted to the Languna Honda Home, in which nonunion resulted in every case. Only five of these cases received primary treatment in this hospital. In 1937, Kleinberg reported a series of 12 cases, treated by a modification of the Whitman method, in which ten, or 83 per cent, were firmly united within two to three months.

In an attempt to determine the reasons for this wide discrepancy, we find several factors which influence the interpretation of the end-results:

(1) The Length of Follow-Up.—Since healing is admittedly slow, and late complications often result, reports based upon a follow-up of two or three months are not comparable to reports of cases followed for one or two years.

(2) Differentiation as to Location and Type of Fracture.—Campbell[9] has pointed out the failure of many authors to differentiate clearly between intertrochanteric and intracapsular fractures, and between impacted and unimpacted

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intracapsular fractures. He[10] has shown that more than 50 per cent of all fractures of the hip are intertrochanteric or impacted. Failure to recognize the variation in prognosis in these various groups leads to confusion in comparing end-results. Magnuson[11] and Smith-Petersen[16] have also emphasized the fact that subcapital fractures, impacted in a valgus position, invariably unite irrespective of the method of treatment employed.

(3) Differentiation Between Recent and Old Fractures.—A report based upon all fractures admitted to a large hospital, regardless of where the primary treatment was received, will of course include many ununited fractures transferred from other institutions or admitted for a reconstruction operation, and will, therefore, result in a high percentage of nonunion.

(4) Statistical Variations.—It is obvious that statistical reports will vary with the author's choice of cases. In comparing end-result statistics it is necessary to know whether the percentage is computed from all cases, including unknown and fatal cases, or from only living, followed-up patients.

In an attempt to determine the results of closed methods of treatment of intracapsular fractures admitted to two hospitals, a study was made of 190 consecutive cases from the New York Hospital over a 20 year period (1915–1935), and from the St. Vincent's Hospital over a ten-year period (1925–1935). Although 190 patients were admitted to these hospitals during the years mentioned, 34 of them were admitted one month or longer after injury, having had previous treatment. Some were admitted for reconstruction operations for nonunion or were treated for some other unassociated illness.

In estimating end-results of the closed methods of treatment, only those cases that received primary treatment in the above hospitals are included. No cases of intertrochanteric fracture are included in this report. The patients were treated by 31 surgeons.

TABLE I

SEX AND AGE INCIDENCE IN 190 CASES OF INTRACAPSULAR FRACTURES OF NECK OF FEMUR

| | |
|---|---|
| *Sex* | |
| Females | 135 cases—71% |
| Males | 55 cases—29% |
| *Age* | |
| Average age | 60 |
| Youngest | 12 |
| Oldest | 95 |
| Cases receiving primary treatment | 156 |
| Cases receiving primary treatment elsewhere | 34 |

*Sex and Age Incidence.*—There were 135 females (71 per cent) and 55 males (29 per cent) in the entire series. The average age was 60, the oldest being 95 and the youngest 12 (Table I).

TABLE II

MORTALITY RATE AND CAUSES OF DEATH

156 Cases Average Age, 72 27 Deaths—Mortality, 17.3%

| | |
|---|---|
| Pneumonia | 8 cases |
| Pulmonary embolism | 8 cases |
| Cardiac failure | 5 cases |
| Multiple injuries | 2 cases |
| Sepsis (from decubitis) | 2 cases |
| Chronic nephritis | 1 case |
| Intestinal obstruction | 1 case |

*Mortality.*—There were 27 deaths, a mortality of 17.3 per cent. It is significant to note that the average age of the fatal cases was 72, 24 of the 27 cases being over 60 years of age. The average duration of treatment before death was 40 days.

A study of the causes of death shows that pneumonia and pulmonary embolism account for over half of the fatalities (Table II). This finding closely parallels the statistics of Howard and Christophe.[6]

In four of the eight cases dying of pulmonary embolism autopsies were performed. In the remaining four the clinical diagnosis seemed justifiable, but no autopsy was done. The high mortality due to pulmonary embolism suggested further study to determine whether prophylaxis or treatment could minimize this complication. The average age of these patients was 70. The average duration of treatment before death was 17 days, the shortest being seven, and the longest 39 days. The form of treatment of the fracture did not seem to bear any relation to these deaths except that all of the patients were bedridden. Four of these cases were treated by the Whitman method and could be turned in bed frequently. Although there was a fairly large number of cases in the entire series in which phlebitis was reported as a complication, in only one of the fatal cases of embolism was this complication recorded.

In view of the high mortality due to pulmonary emboli, it may be of value to consider clotting factors and prophylactic treatment as outlined by Bancroft and Stanley-Brown.[12] The high incidence of pulmonary emboli is also a definite argument in favor of that form of treatment which permits of mobilization and short periods of bed confinement.

TABLE III

RESULTS IN 34 PREVIOUSLY TREATED CASES

| | |
|---|---|
| Deaths | 1 |
| Nonunion | 23 |
| Union | 2 |
| Unfollowed | 8 |

*Results of Treatment.*—The results of cases admitted to the Hospital four weeks or longer after injury are shown in Table III. Of the 23 cases of nonunion, 18 were old cases with frank nonunion at the time of admission. Thirteen of these cases were admitted for, and subjected to, reconstruction

operations, bone-pegging or arthrodesis. The two cases resulting in union were complete midcervical fractures. Both had been treated at home by bed rest only, one for 42 days, and the other for 46 days before coming to the hospital. After admission they were treated by the Whitman abduction method, and both united with absorption of the neck and a coxa vara deformity (Figs. 12 and 13).

TABLE IV

FOLLOW-UP RESULT IN 156 RECENT FRACTURES

| | |
|---|---|
| Deaths (in hospital) | 27 |
| Deaths (within one year of discharge) | 5 |
| Unfollowed | 55 |
| Living cases followed one year | 69 |

CLINICAL RESULTS OF 69 FOLLOWED CASES

| | | |
|---|---|---|
| Satisfactory | 22 | 31.9% |
| Unsatisfactory | 47 | 68.1% |

*Follow-Up Results.*—The follow-up results of the series of 156 recent fractures are shown in Table IV. There were 32 cases known to have died in the hospital or within one year of discharge. Of the remaining 124 cases, 55 were unfollowed, and 69 were reported upon one year or longer after injury. Of these, 46 had had an examination and roentgenograms one year or longer after admission to the hospital and 23 reported by letter. No attempt was made to grade the results as good, fair or bad, in the cases reporting by letter because the information given was too meager to arrive at accurate conclusions. Those cases known to have bony union with good function and those cases reporting stable weight bearing hips, who returned to former occupations, are considered satisfactory. Those cases known to have nonunion with poor function, and those reporting a disability necessitating support in walking, are considered unsatisfactory.

TABLE V

STATISTICS RELATIVE TO RESULTING BONY OR NONUNION IN 46 FOLLOWED CASES

| | |
|---|---|
| Bony union | 8 cases—17.5% |
| Nonunion | 38 cases—82.5% |

METHODS OF TREATMENT IN 46 FOLLOWED CASES

| Procedure | No. of Cases | Nonunion | Bony Union | Percentage of Union |
|---|---|---|---|---|
| Whitman | 35 | 30 | 5 | 14.2 |
| Traction | 9 | 7 | 2 | 22 |
| Sandbags | 2 | 1 | 1 | 50 |

*Bony Union.*—In determining the presence or absence of bony union, only those cases are included in which roentgenologic examination was available one year or longer following the date of injury. Of the entire series of 129 living cases only 46 were examined physically and had roentgenograms taken after

this period of time. This is a low follow-up percentage and illustrates one of the difficulties in compiling an accurate report in such cases. Many are transferred to other hospitals during the immobilization period, many die during the first or second year after the injury and others are totally disabled and cannot return for physical or roentgenologic examination.

Table V shows the results in the living cases followed and examined roentgenologically one year or longer after injury. There are 46 followed cases of which eight, or 17.5, per cent resulted in bony union, while 38, or 82.5 per cent, resulted in nonunion.

Of the 35 cases treated by the Whitman method only five cases (14.2 per cent) obtained a bony union. Of the five successful cases, one was incomplete and three were impacted. Only one of the successful cases was a complete unimpacted fracture.

Adherents of the Whitman method have stated that such poor results indicate that the surgeons did not use the method properly. It seems logical to assume that if, after 30 years of general use, a method is successful only in the hands of a limited number, it should not be advocated as a general method of treatment.

This record of poor results is made more discouraging after a study of the cases which united (Table VI).

TABLE VI

EIGHT CASES OF BONY UNION

| Type of Fracture | No. of Cases | Treatment | Clinical Result |
|---|---|---|---|
| Incomplete | 1 | Whitman | Aseptic necrosis (Whitman reconstruction) |
| Impacted in valgus | 5 | Whitman, 3<br>Traction, 1<br>Sandbags, 1 | Good |
| Unimpacted, complete | 2 | Whitman, 1<br>Russell traction, 1 | Good |

Of the eight cases resulting in bony union, only two were complete, unimpacted, intracapsular fractures, the remaining six being incomplete or subcapital fractures impacted in the valgus position.

*Nonunion.*—In studying the cases of nonunion it was found that, invariably, there was absorption of the neck with more or less upward displacement of the femoral shaft. In some cases the absorption involved not only the neck but also the femoral head. In some cases evidence of nonunion appeared in less than three months but in many instances definite evidence of nonunion was not apparent until after three months. In other cases union was apparently solid, as determined by roentgenologic examination six and nine months following the injury, nonunion being demonstrated thereafter. Upward displacement of the femoral shaft increased after weight bearing was attempted, and in most cases shortening of one or two inches resulted.

Four of the ununited fractures treated by the Whitman method appeared

FIG. 1.—Impacted fracture in valgus, right. Age 32. Whitman treatment. Union. Roentgenogram five years after injury.

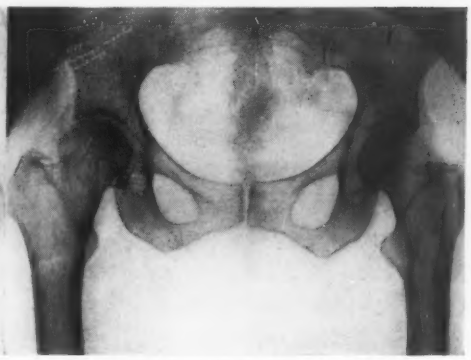

FIG. 2.—Impacted fracture in valgus, right. Age 67. Sand bag treatment. Union. Roentgenogram three years after injury.

FIG. 3.—Impacted fracture in valgus, right. Age 30. Whitman treatment. Union. Roentgenogram ten and one-half years after injury.

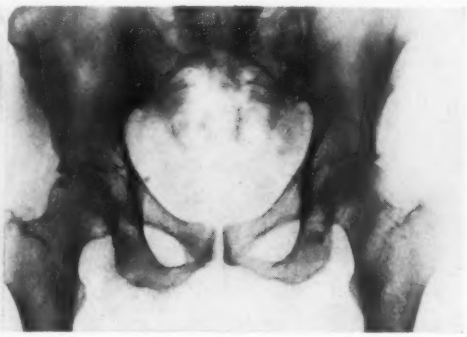

FIG. 4.—Impacted fracture in valgus, left. Age 50. Whitman treatment. Union. Roentgenogram seven and one-half years after injury.

FIG. 5.—Impacted fracture in valgus, left. Age 37. Russell traction. Union. Roentgenogram 14 months after injury.

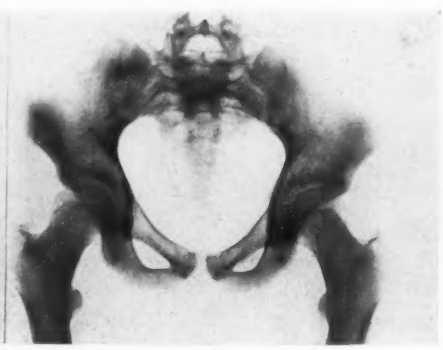

FIG. 6.—Incomplete fracture, left. Age 16. Whitman abduction spica. Roentgenogram at time of injury.

impacted in the original roentgenograms. None of these had lateral views made. All of the remaining cases were complete, unimpacted fractures, most of them with displacement and rotation apparent in the roentgenogram.

Those cases treated in the past few years, since the use of lateral roentgenograms, showed no improvement in results. Many were apparently perfectly reduced in both the anteroposterior and lateral views, yet none of the complete midcervical fractures having the benefit of lateral roentgenograms united.

Eight of 38 cases of nonunion were subjected to operative procedures. Four had Whitman reconstruction operations, one being a failure; the other three were successful. In two cases a Brackett operation was performed, one was successful and one unsuccessful. In one case a bone peg was used with an unsuccessful outcome, and in one case an ostotomy was performed, the result being unknown.

Of the 30 remaining cases, two were able to walk without support and with little pain or limitation of motion, after three years. Both had marked limps but were able to carry on their occupations. Four were able to walk with the aid of a cane, but their locomotion was definitely limited. All of these patients were totally disabled for two or three years. Ten were totally disabled and practically bedridden after a period of three years. The remaining 14 were not followed long enough to estimate function.

*Cases Resulting in Union.*—In the followed cases there were five in which the original roentgenograms showed impacted, subcapital fractures with the head in the valgus position. All of these cases united with excellent anatomic, as well as functional results. Roentgenograms, as shown in Figs. 1, 2, 3, 4, and 5, showed healing with negligible absorption of the neck or deformity of the head except for the valgus position. Three were treated in the Whitman abduction spica, one by a Buck's extension and one by sandbags.

There was only one incomplete fracture in the series (Fig. 6), a boy of 16, immobilized in a Whitman spica for eight weeks. Roentgenograms at regular intervals showed bony union up to one year. Thereafter, the head showed beginning aseptic necrosis, and at the end of three years the process was well advanced in the head and neck with narrowing of the joint space (Fig. 7). Because of pain and disability a Whitman reconstruction operation was recently performed.

This case illustrates the necessity of long follow-up study before end-results may be accurately compiled. It also clearly illustrates that, in injuries to the femoral neck and head, a process of bone absorption, or aseptic necrosis, may result without a complete fracture. There is much emphasis in the literature upon accurate apposition and adequate fixation as necessary factors in the prevention of nonunion, but in the above case these requirements were necessarily fulfilled. The incomplete fracture united. Nevertheless, aseptic necrosis developed. The cause of late changes in the femoral neck and head is not known, but undoubtedly it has to do with injury to the cartilage of the head and to disturbance of blood supply of the head and neck. Speed[13] has emphasized the fact that this process of absorption may proceed in spite of

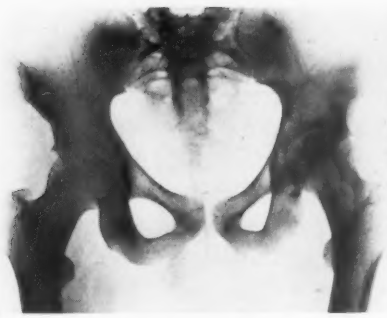

FIG. 7.—Roentgenogram after three years. Fracture united. Aseptic necrosis of the head.

FIG. 8.—Roentgenogram after reduction of dislocation of right hip.

FIG. 9.—Roentgenogram after three years showing aseptic necrosis of the head and neck.

FIG. 10.—Unimpacted fracture. Age 70. Russell traction. Union. Roentgenogram two years after injury.

FIG. 11.—Unimpacted fracture, right. Age 56. Whitman treatment. Union. Roentgenogram two and one-half years after injury.

FIG. 12.—Unimpacted fracture, left. Age 60. Admitted 42 days after injury. Whitman treatment. Union. Roentgenogram 15 months after injury.

accurate fixation of femoral neck fractures. The same process may follow dislocation of the hip without fracture of the femoral head or neck. Figure 8 shows a roentgenogram of a right hip after closed reduction of a traumatic dislocation. There was a fracture of the rim of the acetabulum. After three years, symptoms of pain and shortening occurred and a roentgenogram (Fig. 9), revealed aseptic necrosis of the head and neck. From a study of these cases it is evident that a process of absorption of the neck or head may occur when the problems of proper reduction and immobilization are eliminated. This fact may explain the frequent occurrence of nonunion in midcervical fractures.

Only four complete midcervical fractures were found to have united in the entire series, two being fresh fractures. One was treated by Russell traction, because the age (70) and general condition of the patient contraindicated more radical treatment. Figure 10 shows the roentgenogram of this case, two years after injury with definite bony union and no apparent shortening of the neck. The other recent case was treated by the Whitman method and the result, Fig. 11, two and one-half years after injury, shows bony union with absorption of the neck and a coxa vara deformity. The functional result in this case is very good. The remaining two midcervical fractures were admitted for Whitman treatment 42 and 46 days, respectively, after injury. Both united with absorption of the neck and a coxa vara deformity (Figs. 12 and 13), but with good function.

In practically all midcervical fractures of the neck of the femur, absorption of the neck occurred and union rarely resulted. The results of the cases shown in Figs. 11, 12, and 13 are unusual in this series. In spite of absorption of the neck, the head has united to the shaft. These results are similar to that obtained by Magnuson[14] in his adaptation of the Brackett operation to fresh complete fractures (Fig. 14). In this operation a portion of the neck which usually absorbs is sacrificed in reforming the upper end of the fractured neck to be inserted into the hollowed-out head. The greater trochanter is transplanted to a lower position on the shaft. Although this operation is a formidable one, and is applicable only to those patients in fairly good condition, the theory of the procedure is sound, and Magnuson[11] has reported satisfactory results.

Investigation of the complete intracapsular fractures in this series of cases would indicate that closed methods of treatment are still inadequate.

With modern internal fixation[2, 17] these elderly patients are able to be out of bed and may bear weight on the affected leg even before union can occur. These methods are a marked improvement over closed methods of treatment in this respect. Smith-Petersen states that the nail, in itself, does not promote union but allows early mobilization with firm fixation. The method has not been employed over a long enough period to establish the end-results in respect to bony union, but even if late reports indicate a low percentage of bony union, the internal fixation is still an improvement over the closed methods.

Since absorption of the neck with nonunion is the usual result in unim-

pacted intracapsular fractures, it seems that exponents of the early reconstruction operations have evidence upon which to base their plan of treatment. In these cases, if operative procedure may be safely performed, it seems wise to accept inevitable nonunion and to undertake early operative procedures for reconstruction, such as the Magnuson-Brackett[14] operation, or the Schanz osteotomy as described by Schumm.[15]

FIG. 13.—Unimpacted fracture, left. Age 39. Admitted 46 days after injury. Whitman treatment. Union. Roentgenogram 15 months after injury.

FIG. 14.—Brackett operation for fresh, complete, unimpacted fracture (from Magnuson). Roentgenogram two years after operation.

## CONCLUSIONS

(1) Accurate end-results of any method of treatment should differentiate between: (1) Intracapsular and intertrochanteric fractures; and (2) impacted and unimpacted fractures.

(2) Accurate end-results of treatment with respect to bony union cannot be obtained without at least one year follow-up roentgenologic examination.

(3) Accurate end-results concerning function cannot be obtained under one or two years because: (1) Some cases resulting in bony union may develop aseptic necrosis of the head and neck; and (2) some cases resulting in nonunion have a fairly satisfactory functional result.

(4) In this series, subcapital fractures impacted in the valgus position, united with little or no deformity of head or neck, regardless of the types of treatment.

(5) Complete unimpacted fractures usually result in nonunion with absorption of the neck.

(6) Absorption of neck and head may develop even with apposition of fragments and proper immobilization.

(7) In this series of cases the Whitman method has shown too few satisfactory results to justify its continued use.

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DISCUSSION.—DR. JAMES M. HITZROT (New York) was greatly interested in hearing a discussion on what Kellogg Speed has called "the unsolved fracture," and what Cotton has recently described as the fracture for which the best procedure has not yet been discovered; namely, intracapsular fracture of the neck of the femur. The essential result desired is a satisfactory weight-bearing leg, without pain, and with sufficient motion at the hip joint to allow the patient to sit down with comfort. A great many methods have been devised and are at present being employed to accomplish this result. Just how satisfactory many of them are is a matter open for discussion.

Doctor Hitzrot considered intracapsular fracture of the neck of the femur as occurring in three age-groups: The first is in young people, before the epiphysis of the head of the femur has united to the shaft; the second, in patients between the ages of 20 and 50; and the third, in patients of 50 years of age or more. The problems of treatment are a little different for each

group. In the first group, fractures of the neck of the femur or epiphyseal separations require complete, early reduction, and unless this is obtained satisfactorily by manipulation these cases should be operated upon early and the replacement obtained by actual exposure of the fracture line, every attempt being made to conserve the growing portion of the bone. In the second group, reduction by open operation or by fixation with various forms of metal nails, screws, etc., is probably the method of selection, and, in Doctor Hitzrot's opinion, the future will see more operative procedures in this group than at present. In the group over age 50, and especially in the older members of this group, the primary essential of treatment is the general care of the patient because most fatalities occur in this group. Naturally, any series of cases from age 60 upward, will show the highest mortality, and in these elderly people that type of treatment must be instituted which will give them the most comfort and run the least risk of endangering their lives. The incident of nonunion in patients more than 50 years of age, is highest. Therefore, the results in this group are as yet not satisfactory in that, after a long period of treatment, these patients usually have an unsatisfactory weight-bearing hip. This occurs no matter what the original treatment may have been, since the etiologic factors and cause of the nonunion are not controlled by any of the operative procedures. That the newer methods of fixation by nails, screws, etc., are a more comfortable treatment, allow these patients more freedom, and are less confining, can generally be accepted, but the late results are not as yet proven to be more satisfactory than those of the earlier manipulative methods. If nonunion occurs due to absorption along the fracture line the same failure to obtain a satisfactory weight-bearing leg results.

Fractures of the neck of the femur deserve consideration also from another point of view; namely, some type of operative procedure which will avoid the disappointing results and which will enable the patient to be up and about with a weight-bearing leg and a minimum amount, if not an entire absence, of pain on walking.

Doctor Hitzrot felt that, in properly selected cases, some type of reconstruction operation performed early would give the patient a more satisfactory weight-bearing leg than could be obtained by any form of treatment which attempted to obtain union along the fracture line. In a discussion with Doctor Magnusson, Doctor Hitzrot understood him to be of the opinion that the Brackett operation, performed early, would produce this result. Even if union does not occur following this operation, the weight-thrust will give the patient a satisfactory weight-bearing leg without pain, and the patient will be able to get about earlier than if operation were undertaken after nonunion has occurred.

Doctor Hitzrot emphasized that an operation, of the type described by Whitman, the so-called "Whitman reconstruction" operation, must also be given consideration. If this is undertaken early, the resulting weight-bearing leg is much more satisfactory than if it is performed late. Doctor Hitzrot's experience with 20 cases, in which this procedure was carried out, after it was definitely established that nonunion was occurring, demonstrated satisfactory results in 18 instances, which led him to the conclusion that if the operation had been performed early the result would have saved the patient a good deal of trouble. His experience with other types of procedure or the results which he has seen have not seemed entirely satisfactory. His results with the Brackett operation have not been quite as satisfactory as those obtained by Doctor Magnusson. This, he thought, might be due to

a fault in his operative technic, in that he did not get the head fragment so fixed that the weight-thrust was directly upward through the shaft, and when the weight-thrust becomes exerted at an angle as the result of this operation the end-results are likely to be disappointing.

Neither of these operations is a minor undertaking, and the cases upon which they are performed should be carefully selected. If the patient is properly prepared, Doctor Hitzrot's experience with the Whitman reconstruction operation has been such that in his opinion many of these patients will stand one of these operative procedures without any great difficulty. Just what the mortality will be in these cases is problematic. The cases upon which Doctor Hitzrot had operated late have, up to the present, all been carefully prepared and selected for the operation. Among these he, fortunately, has not had a fatality. If this care in selection is not made, naturally the mortality will be greater.

Doctor Hitzrot's experience with the various types of bone grafting in these fractures, which he began to employ as early as 1912, using autogenous and beef-bone dowels and screws, had not given satisfaction, and when healing occurred in the early cases it seemed that it would have occurred equally as well without the operation. There is nothing about the bone graft that will alter the process which tends to produce absorption along the neck, even in young individuals, and the fixation obtained by these various types of graft has not been as firm as that which can be obtained by various types of metal nails and screws, and is, in addition, much more difficult of accomplishment.

Although reconstruction operations leave the patient with a shortened leg, they do not result in much more shortening than occurs when the neck bends later, nor do they give as much shortening as occurs after the absorption has taken place on the shaft side of the neck. The early application of weight to the reconstructed neck prevents, to a large degree, this absorption, and Doctor Hitzrot was convinced that some such radical procedure as the Brackett operation or the Whitman reconstruction operation must be given due consideration in the treatment of these unsolved fractures. Just what procedure will give the best result will have to await the results obtained by many well equipped surgeons, and to obtain a true perspective on these results, the cases should be followed for at least two years or longer after injury before arriving at any conclusion as to the desirability of either procedure.

Dr. Donald Gordon (New York) reviewed Doctor Wade's statistics in seven series of end-results for the Whitman method, which varied from 53 to 83 per cent satisfactory. If the figures of Whitman are excluded, the average of successful results was 55 per cent, including Mensor and Dewey's statistics of 50 cases with zero success. If that group is left out, the average becomes 66.2 per cent, which compares favorably with recent reports of nearly 70 per cent in series analyzed by other men. Mensor and Dewey's figures depart so far from the major cross-section of the results quoted by Doctor Wade, that it would appear that the surgeons made such a poor application of the Whitman method that it is questionable if they would have done any better by employing a more complicated procedure.

Concerning Doctor Wade's statement with regard to the group he studied—that "It seems logical to assume that if after 30 years of general use a method is successful only in the hands of a limited number, it should not be advocated as a general method of treatment"—Doctor Gordon said that the statistics shown by Doctor Wade for the two New York hospitals certainly reflected little credit on the method or the hospitals, as compared with those quoted by men from other cities, the general reports of success with the Whitman method

as to end-results, whether functional or with bony union, being at variance with the figures shown in Doctor Wade's study. This study did not reveal any data to suggest a technic which would afford any better results in the hands of those unable to use the Whitman or closed methods with an average degree of success. Doctor Gordon was not convinced from the data presented that closed methods of treatment should be given up. Human judgment suffers a severe strain in choosing which method of procedure to employ where a study of Table V, presented by Doctor Wade, reveals that two cases treated with sandbags gave a 50 per cent incidence of bony union in the series of cases critically examined by Doctor Wade.

Discussing the ingenious and specialized technic presented by Doctor Bergamini and Doctor Dooley, Doctor Gordon thought that their own, combined with other standard procedures, involved the introduction of a pin of Vitallium with a shearing resistance up to 24,000 pounds, or 12 tons, and said that such a pin should resist corrosion. Its prolonged tolerance by the tissues of various patients will have to be learned. Its strength should support weight-bearing of even the very obese, until the fracture has had an opportunity to unite, if it will, or for a longer period if it will not.

The method of finding the angles formed by the axis of the neck and shaft from the tracing is simple and useful, but it would be of help if the authors would state what corrections should be made when the upper end of the femur is not a normal one, because of former pathology. The control of the direction of the pin appears to be within the safety limits required. Doctor Gordon wondered if the authors had had any pins deviate enough to cause a complication.

Doctor Gordon stated that the Leadbetter[1] method of reduction stood alone in reducing and proving reduction before being controlled roentgenographically. He felt that the technic of the lateral view of the normal or injured hip, as shown by Doctor Bergamini, was most novel and should be helpful for other purposes in this region as well as for localizing foreign bodies other than the pin. The determination of the precise position of the injured or well leg by the use of the inclinometer would be precluded where a stiff knee or mid-thigh amputation existed in either leg, and, therefore, a simpler technic would be necessary.

Only one stage in the development of the technic for the introduction of flanged pins in treating a fracture of the neck of the femur was presented. Conclusions were furnished based upon the authors' extensive personal experience, which Doctor Gordon found it difficult to agree with completely, mainly on the basis of his lack of similar experience with an entirely new procedure, and in the absence of any data on their results. The time allotted for the presentation, of course, did not permit the presentation of these data; without it one would not be in a position to apply the yardstick of the results of similar pinning procedures or to pass judgment on its comparative value. Certainly the attention to the many details which appear to furnish precise methods in each step, leaving little to fallacious judgment, would appear to render a better operative result and afford the surgeon the satisfaction of precision which should eliminate one of his worries.

Doctor Gordon doubted whether the skillful technical minds of the authors would remain satisfied even with the present technic, and believed that they would simplify it in the future. The procedure appears to give a great degree of accuracy in locating where the pin is to be placed and in directing its course, but the authors, while assuring that the procedure is simple, do not mention how many hours must be spent on the development of an organization, in

order to enable one to carry out the method simply. This encouragement is needed to induce a trial of any method; but it is only too obvious that anyone attempting the procedure presented, without first-hand experience with those who have developed it to its present degree of perfection, would find many pitfalls. It is the early errors, produced by the pitfalls resulting from inexperience, which tend to detract from the merits of a technic in its early life, and to obscure its true value in the late results. A report in the future by the authors of their complications, sequelae and three-year results based on present standards will be of great value in determining and preserving the good points of their technic. In the main problem, when any pin or internal retention device has been placed in a reduced fracture, any technic must be submitted to the many as yet unknown factors of physiology and pathology and the limited life expectancy in cases of this "unsolved" problem.

Doctor Gordon said that, although inexperienced in pin work, he personally believed that in the hands of a properly trained organization it affords accurate reposition. A small, strong, noncorrosive pin should cause a minimum of damage in the open operation which such work requires. Doctor Bergamini's and Doctor Dooley's method appealed to the speaker as a step forward in pinning technic which, by early mobilization of the patient to the upright position and by improving the blood supply of the femoral neck, will do much toward bringing about a union and minimizing the subsequent bone absorption.

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Dr. Paul K. Sauer (New York): Without minimizing in the slightest the credit due Doctor Whitman, it must nevertheless be admitted, as has been so well demonstrated by Doctor Wade, that the Whitman method except possibly in the hands of a select few has not proved adequate in the treatment of intracapsular fractures of the neck of the femur.

During the past six years, at the City Hospital, Doctor Sauer had remarked the continued readmission of fracture of the hip cases year after year, which gave rise to the suspicion that their method of treating intracapsular fractures of the femoral neck left much to be desired. Their impression of the poor results was not only substantiated, but upon analysis of the cases from 1929 to 1936, it was ascertained that the actual results were even less favorable than they had suspected.

In Doctor Sauer's opinion no result that is less than two years old should be considered, much less reported, as a careful follow-up of results at the end of two or more years often dispels the impression of a bony union, which had seemed a successful accomplishment at the end of nine months or a year.

During the seven years up to January, 1936, there were 96 cases of intracapsular fractures of the neck of the femur treated at the City Hospital. (No cases of intertrochanteric fractures are here included.) It was noted that although several cases were reported by the roentgenologist as being impacted when they were first examined roentgenologically, not a single case of impaction was found on later examination.

An analysis of the 96 cases showed the sex incidence to be 63 females and 33 males, a ratio of approximately 2:1. The average age of all the patients was 62.5 years; the youngest 26, and the oldest 95. The mortality rate was 25 per cent.

TABLE I

ANALYSIS OF 96 CASES OF INTRACAPSULAR FRACTURE OF THE NECK OF THE FEMUR

| | |
|---|---|
| Total number of cases | 96 |
| Average age | 62.5 |
| Youngest | 26 |
| Oldest | 95 |
| Average morbidity | 5.3 mos. |
| Mortality | 25% |
| Cases receiving more than one method of treatment, other than reconstruction operation | 27 (35%) |
| Number of cases of bony union | 0 |
| Number of cases receiving reconstruction operations | 19 (20%) |

Table II shows the various methods of treatment instituted. A justifiable question would be how many of these cases were properly reduced in the first place before the application of the Whitman spica, and in how many of those properly reduced was the reduction maintained after the application of the spica? I don't know; but I assume just about as many as are usually reduced and maintained in the average hospital. You will also notice in Table II that the only case in which bony union was obtained, was complicated by a simultaneous intertrochanteric fracture in which a long upper fragment was wired to the shaft. Otherwise the record of bony union is 100 per cent nil. Twenty-seven, or 35 per cent, of these cases received more than one method of treatment and still there was no bony union obtained.

TABLE II

ANALYSIS SHOWING METHODS OF TREATMENT EMPLOYED AND RESULTS

| Methods of Treatment Employed | No. | Average Age | Age of Youngest | Age of Oldest | Average Morbidity | Mortality | Mortality Per Cent | Union | No. of Cases Receiving Other Treatment |
|---|---|---|---|---|---|---|---|---|---|
| Unknown | 4 | 59 | 43 | 76 | Unknown | 1 | 25 | 0 | 0 |
| Russell traction | 6 | 58 | 34 | 76 | 1 mo. | 0 | 0 | 0 | 5 (83%) |
| Whitman spica | 58 | 63.5 | 26 | 86 | 4.8 mos. | 8 | 14 | 0 | 15 (21%) |
| Bozsan drilling; with spica | 8 | 65 | 54 | 79 | 4.9 mos. | 1 | 12.5 | 0 | 4 (50%) |
| Bozsan drilling; Roger Anderson | 1 | 34 | 34 | 34 | 3 mos. | 0 | 0 | 0 | 0 |
| No treatment; sandbags | 12 | 69 | 53 | 95 | 19 mos. | 6 | 50 | 0 | 1 (8%) |
| Roger Anderson; wiring shaft | 1 | 45 | 45 | 45 | 7.5 mos. | 0 | 0 | 1* | 0 |
| Kirschner wire; pinning | 3 | 63 | 56 | 70 | 5 mos. | 1 | 33 | 0 | 1 (33%) |
| Jones splint; bone graft | 1 | 42 | 42 | 42 | 9 mos. | 0 | 0 | 0 | 1 (100%) |
| Thomas splint; walking caliper | 2 | 65.5 | 55 | 76 | 1.5 mos. | 1 | 50 | 0 | 0 |
| Smith-Petersen nail | 1 | 40 | 40 | 40 | 5 mos. | 0 | 0 | 0 | 0 |
| Moore nail | 1 | 76 | 76 | 76 | 1.5 mos. | 1 | 100 | 0 | 0 |
| Wire nail | 1 | 26 | 26 | 26 | 5 mos. | 0 | 0 | 0 | 0 |
| Buck's extension | 1 | 67 | 67 | 67 | 2 mos. | 0 | 0 | 0 | 0 |

* Bony union obtained; fracture complicated, however, by an intertrochanteric fracture, which was wired.

Based on the findings of our analysis of these cases it was decided that we were most unsuccessful in reestablishing these people as an economic asset to the community, and that our usual method of treating these cases was faulty. It seemed that if nonunion was to be the rule in these cases when treated by

the ordinary methods, some other type of therapy had to be instituted. Therefore, it appeared that some type of reconstruction operation as the primary treatment was indicated in order to obtain an early satisfactory result.

TABLE III

ANALYSIS OF THE 19 CASES WHICH RECEIVED RECONSTRUCTION OPERATIONS

| | |
|---|---|
| Number of cases having reconstruction operations | 19 |
| Average age | 43.3 |
| Youngest | 28 |
| Oldest | 76 |
| Number having had previous treatments | 18 |
| Average morbidity before reconstruction | 7 mos. |
| Average morbidity after reconstruction operation | 4½ mos. |
| Mortality (4 cases) | 21% |
| Results: | |
| Good | 11 |
| Fair | 1 |
| Failure | 3 |

TABLE IV

ANALYSIS OF THE TYPES OF RECONSTRUCTION OPERATIONS EMPLOYED AND RESULTS

| Types of Reconstruction Operations | No. | Average Age | Age of Youngest | Age of Oldest | No. Previously Treated | Average Morbidity before Reconst. | Average Morbidity after Reconst. | Mortality | Mortality Per Cent | Results |
|---|---|---|---|---|---|---|---|---|---|---|
| Whitman reconstruction | 3 | 56.3 | 52 | 64 | 3 | 10 mos. | 7 mos. | 2 | 66.6 | 1 walking |
| Brackett operation | 1 | 28 | 28 | 28 | 1 | 3 mos. | 5 mos. | 0 | 0 | Ankylosis, walking |
| Lorenz bifurcation | 15 | 60.6 | 39 | 76 | 14 | 7.7 mos. | 4½ mos. | 2 | 13.3 | 9 good<br>1 fair<br>3 failures |

Four types of operation were considered: Whitman reconstruction, Colonna's operation, Brackett's operation, and the Lorenz bifurcation. The first two were discarded because they necessitate opening the hip joint and removing the head of the femur. Frequently these operations lead to ankylosis of the joint. The Brackett operation, while it makes use of the head, demands that the joint be opened, a procedure which is not without the attendant dangers of shock and infection. Therefore, the Lorenz bifurcation was chosen as being the most suitable to be employed at the City Hospital for the following reasons:

(1) The hip joint need not be opened, the operation being an extracapsular osteotomy.

(2) The head of the femur is left in place, and is used as a movable point of support.

(3) There is practically no shock to the operation, and the procedure can be borne by any patient capable of undergoing an operation.

(4) The time of operation is short, 20 minutes being all the time required except for the application of the plaster encasement.

(5) The danger of infection is minimal. If infection does take place, the extracapsular tissues are much more capable of taking care of it than the joint surfaces.

(6) There is very little shortening of the leg. While there is some absolute

shortening, there is relative lengthening due to the abducted position of the leg and the tilt of the pelvis when walking. However, a heavy sole on the shoe of the affected leg will overcome most of the limp.

(7) The mortality is very low. Out of our series of 15 cases there were only two deaths. One patient died of a coronary occlusion ten days after operation, and the other one of a pneumonia two and one-half months post-operative.

There is, to-day, no one satisfactory method of treatment that is applicable in all cases. Many factors, such as the economic and physical status, age, and the conditions under which treatment must be rendered, will of necessity compel different methods of treatment to be instituted.

Dr. William Darrach (New York) also felt, as had been mentioned by the speaker, that it was a great mistake to draw any conclusions relative to bony union or the vitality of the femoral head during the first two years, and cited three cases illustrating this contention:

The first case was that of a woman, age 72, who had had an open reduction with the introduction of a Smith-Petersen pin. She was walking around at the end of nine weeks. She continued to walk without pain and with almost complete motion until the thirteenth month. At this time while turning over in bed she experienced a sudden severe pain in her hip and was unable to bear her weight on it. Roentgenologic examination showed that the pin had broken and that the fragments had separated. At the end of seven years there was almost complete absorption of the head.

Another case showed bony union nine months after open pinning, with good motion. The pin was removed, and her range of motion gradually decreased and the pain increased. Roentgenologic examination showed marked flattening of the head, notwithstanding the fact that bony union had occurred.

The third case, a woman, age 29, had come up from the South in a plaster spica with an end-to-side position of the fragments. A closed reduction seemed to give good apposition, but after five months it was evident that there was no union and an open reduction was carried out with insertion of a pin. This also failed and a second open reduction and reinsertion of a pin was carried out 11 months later. This resulted in bony union, but after three years the head had become considerably flattened, and at the end of seven years a roentgenogram shows a very misshapen, flattened head. Yet in spite of this there is evidence of a good joint space. She has a marked limp, marked limitation of abduction, internal rotation and flexion, but often spends most of the day on her feet, rides horseback and does not use either a cane or a crutch.

These three cases demonstrate that one cannot always tell at the end of one or even two years, what the results will eventually be. The problem of intracapsular fracture is still "unsolved." With regard to trochanteric fractures, one does not have to worry quite so much. However, a great deal of encouragement may be found because of the marked improvement which has taken place in the treatment of these cases. A few years ago, C. R. Murray made the statement that: To get good union one should first get a good reduction; second, maintain that good reduction; and third, get bony union. Doctor Darrach added two other factors: (1) You have to save the patient's life; and (2), you have to protect in some way, if possible, against the late changes that tend to take place, such as flattening out of the head, breaking down or shortening of the neck. These five points must all be borne in mind.

Everybody's mortality has been reduced a great deal—Doctor Darrach said this was true of his because of a number of factors in this special group, one of which he thought was internal fixation. His attitude at first had been that

internal fixation, that is, open reduction with insertion of a Smith-Petersen pin, should be performed in all cases that could withstand such a procedure. However, he now feels that an open reduction and employing a Smith-Petersen pin should be performed only if it is thought the patient will not stand plaster encasement and long bed stay. The pendulum has swung the other way. In older people, open reduction seems safer than to employ a plaster encasement, even though it is a long operation. Doctor Darrach's mortality had been definitely reduced both by undertaking an open operation, and through better after-care. A great many more lives can be saved if the patient can be enabled to get up sooner. The position on this subject seems to change every two or three years, and it has to do so if anything is to be learned. Doctor Darrach said he was not certain what is the best method of handling these cases. One series was tried employing the Smith-Petersen procedure, not dividing the glutei; and then another series in which, in order to improve the blood supply, some muscle was transplanted to the head fragment, but the results did not compare favorably with those of some others, especially those reported by Thornton, who has had 60 cases, in which he has obtained most excellent results from blind nailing. Doctor Darrach said he was now going to start in doing some blind nailing. He did not think, however, that he was up to using the fascinating arrangement described by Doctor Bergamini.

One must keep trying out different methods, studying results, not deceiving one's self, following one's cases through, and actually finding out just what the conditions are at the end of two, three or five years. If all will do this, then, in another 15 or 20 years, it will probably be possible to get somewhere in solving this most difficult but most fascinating problem; namely, fracture of the neck of the femur in the capsule where the blood supply is poor.

Dr. Frederic W. Bancroft (New York) said that it had been his good fortune to have heard many talks on fractures of the neck of the femur during the period of three years in which he has been connected with the Committee on Fractures, so that he has a passive knowledge without great actual experience on the subject. On the other hand, he said he was not a strong protagonist of one method or the other, and had formed some theoretic ideas from listening to discussions. He thought everyone would agree that the four steps in the progress of treatment of fractures of the neck of the femur were:

(1) The Whitman method, which produced the first real active principle of abduction in the treatment of fractures of the femur.

(2) The introduction of the lateral roentgenographic view, which showed that the anterioposteror view did not adequately give the position of the fractured ends and showed that a great many femurs treated by abduction had never been properly reduced.

(3) The Leadbetter or some similar method, which presented the actual method of reducing the fragments.

(4) Internal fixation.

Doctor Bancroft was sure, that if he received a fracture of the neck of the femur he would wish first to be in the hands of a man well versed in the treatment of fractures. He was also sure that he would wish some means of internal fixation. He also felt that he should like the method employed by Doctor Thornton, who treats a fracture of the neck of the femur as an immediate emergency operation. If admitted at night the patient is treated as any other emergency would be. This procedure has a very definite advantage: The fracture is reduced with a minimum amount of trauma, before the capsule is distended with fluid, and before the surrounding muscles have become

infiltrated with blood. If the reduction has been delayed for several days, fibrin must have adhered to the ends of the bone, as is seen in cases where open reduction for fracture of the patella is performed, and good approximation cannot be effected between the ends of the bone if they are coated with fibrin as can be accomplished when they are not.

The problem of the type of internal fixation is still to be determined. Doctor Bancroft said his preference would be Moore nails rather than the Smith-Petersen nail, as they seem to give a better fixation of the fragments with less foreign body reaction. The ideal has not been reached because, as Doctor Wade showed, even with internal fixation the neck may become absorbed, resulting in nonunion. In one of the roentgenograms shown, where the bone was presumably being extruded, it seemed to him that the extrusion was the result of the shortening of the neck and not eviction of the nail. If, on the other hand, Doctor Bancroft should not be in a hospital, where immediate operation could be performed, and delayed union resulted, he would like to have a reconstruction operation, probably based on the Brackett procedure. This assumed, he said, that he would be in a hospital with proper facilities for roentgenograms to be taken in the operating room, and with an adequate method for taking a lateral view.

Dr. Armitage Whitman (New York) agreed with almost all of the speakers' statements, and appreciated his very painstaking follow-up of the routine cases as they occurred in two hospitals over a period of 30 years. However, statistics were so often deceptive. One had but to refer to the December, 1937, issue of the Journal of Bone and Joint Surgery which contained three articles on fracture of the neck of the femur, and which would seem to reflect the latest expression of authoritative opinion. One of them was written by Doctor Leadbetter, presenting statistics from 1913 to 1937. The best series was one of seven cases reported by Doctor Henderson, who employed a nail, and who obtained 100 per cent union. The next best was Albee's series, who obtained 97.5 per cent union after using a tibial graft. Stern, with the abduction method, obtained 87 per cent union, and so on down to Doctor Katzenstein who had 11.5 per cent union. The British Fracture Commission, before the abduction method was instituted, reported 13.8 per cent. None of the series descended to the depressing results reported by Doctor Sauer. Doctor Whitman said his father has been very much criticized, because he himself had not reported a series of his own end-results. As a practicing orthopedic surgeon at the Hospital for the Ruptured and Crippled he saw only a very small percentage of fresh fractures. With regard to the remark that was made to the effect that Dr. Royal Whitman was responsible, in 1904, for bringing this fracture out of the limbo of complete neglect to the cynosure of surgical attention, Dr. Armitage Whitman felt that it was quite a feat, and he considered that if his father were to die tomorrow he would be satisfied with what he had done. Certainly anyone treating cases of fracture of the shaft of the femur would be criticized if he gave no idea of when the fracture was received or treatment instituted.

One discussor brought out a very good point; namely, that Doctor Thornton gets up in the middle of the night to treat these fractures as one would do if they were any other fracture. In an article by Doctor Harris, in the December, 1937, issue of the Journal of Bone and Joint Surgery, a series is reported, in which 76 per cent bony union was obtained by the nailing operation. Harris says that about two years ago Dr. Kellogg Speed referred to this fracture as the "unsolved" fracture. Harris says that simply by using the methods of treatment accorded any other fracture—namely, immediate accurate reposition, prolonged immobilization, and prevention of weight-bearing until such

time as the fracture has had time to consolidate which, in these cases being oblique, would be much longer than in any other—fractures of the neck of the femur would have as good opportunity of resulting in bony union as any other fractures in the body. Doctor Harris is not concerned as to whether or not the head undergoes necrosis. He has observed cases for more than three years in which this has taken place but in whom immobilization was kept up by the nail, and in which the fracture had, nevertheless gone on to complete healing.

Doctor Whitman could not believe that Doctor Wade was as pessimistic as he sounded when he remarked that: "The Whitman method has shown too few satisfactory results to justify its continued use." The abduction method represents an attempt to treat a fracture, in contrast to the complete neglect in which that fracture lay at the time the abduction treatment was conceived. There can never be any question of the abandonment of the abduction method. It is proper to emphasize that the abduction method is a procedure which results in an accurate reduction of the fracture. Whether the subsequent fixation be by external means, such as a plaster of paris encasement, or by the insertion of a nail, or bone graft, is a detail which must always rest at the discretion or skill of the individual who happens to be treating a given patient.

Doctor Whitman was surprised that one of the discussers had gone back to the text-book dictum that the "first indication is to save life." He hoped that in surgical judgment surgeons had proceeded further than that. Is it not taken for granted, he asked, that any one called in to treat a patient would have as his first indication the saving of life? Abandoning statistics, Doctor Whitman concluded his discussion with the following allegory:

He had had the good fortune of serving three of his six months of medical service at The Roosevelt Hospital under Dr. Frank Jackson, a disciplinarian of the old school. A case of empyema had applied for admission to the hospital. The house physician was so sure that the patient had empyema that he turned him over to the surgeons without waiting for Doctor Jackson. Somebody suggested needling him before opening the chest. Doctor Darrach promptly said: "I will," and needled him several times without obtaining pus. There were two or three cases of carcinoma of the pancreas or something equally grave waiting, and so he sent the empyema patient back to the medical service. No sooner did he arrive there than Dr. Evan Evans came in, at the head of a party of students, looking for something to demonstrate. He lit on that poor man.

Said Doctor Evans, "Why, of course he has empyema!" "The surgeons have needled him," he was informed, "and could not find any pus." "Oh, the surgeons needled him, did they? Well, give *me* a needle!" Doctor Evans tapped the patient on the back several times, with all the students standing around him, listening and watching. "It's a classical case of empyema," Doctor Evans emphasized. Confronted with a case like that, Doctor Evans was like a fox terrier after a woodchuck. All considerations of local anesthesia were abandoned. He needled this way and that way, until, at one o'clock, fortunately for the patient, the lunch bell rang. Doctor Evans and his students disappeared. Well, the poor patient looked like a strawberry cushion Mother used to stick pins in, and was sent back to his bed more dead than alive.

At two o'clock Doctor Jackson came in again. "Any new cases, Barnum?" he asked. "Yes, a case we thought had empyema." "What do you mean, you *thought* he had empyema? Either he has or he hasn't." He whacked the patient on the chest and then put his head down. He never used a stethoscope. He used to listen to the chest through a sheet and his beard, and no one ever could find out how he could hear through his beard. "Empyema? Of course he has empyema . . . bring me a needle!"

"But, Doctor Jackson," protested Barnum, "this man was needled by the surgeons and then Dr. Evan Evans spent two hours needling him." "Barnum, did you hear what I said? Bring me that needle!"

It was like Sidney Franklin spearing a bull blindfolded. Barnum brought the needle. Doctor Jackson thrust it in, and pulling out the plunger, obtained a syringe full of thick, creamy pus. "There you are, Barnum," he said quietly. "That will show you it makes some difference who does the needling. Next case."

DR. W. RUSSELL MACAUSLAND (Boston) said that up to the time when Dr. Armitage Whitman spoke he had developed several good ideas but then they vanished. It must be a comfort to many to know that Dr. Armitage Whitman ever served under any period of strict discipline. He could not agree with the statement that statistics mean nothing, and could not appreciate how anyone could formulate a single didactic method of treatment of this complicated fracture, varying as it does in different ages and under very differing circumstances, between the ages of 50 and 90.

It had certainly been a great pleasure to hear so many references to Dr. Royal Whitman's work, for he had served under him and had held him in the highest esteem for 30 years. He so constantly preached the principles of joint care that no surgeon dealing with bones and joints today can practice without these principles, and these principles will live forever—in memory of his clear thinking and sane judgment. The statistics from the Mayo Clinic on the abduction method of treatment of fracture of the neck of the femur, as well as those of Campbell and a series of several hundred from his own clinic, are well within 1 per cent of each other as regards obtaining good bony union, which statistics were scrutinized by unbiased persons; a true evaluation, therefore, of the abduction method would appear to have been obtained. Doctor MacAusland wished that he could feel that the statistics reported relative to the nailing method were of equal valuation. Many surgeons of today, when young, were enthusiastic advocates of the open treatment of fracture. Time has mellowed that, and one must first consider the type of patient and the possible complications. He must consider the surgical technic of the operator and the institution in which this surgery is being performed, and his understanding of that particular fracture in that particular person. Certainly no law can be laid down on any classified or routine method of treatment of this fracture, other than that it first requires reduction. If one is an advocate of the open method, there are certain complications that may follow that. Many must remember the visit of Lane to this country, when metal plates were advocated in most fractures, following which for four or five years osteomyelitis had to be treated, at least all over New England. Doctor MacAusland said he had seen a lot of nailing of these fractures; indeed, some pretty good ones were just presented by Doctor Wade and by Doctor Bergamini, both of whom Doctor MacAusland wished to congratulate on their very sane presentation of the subject, but, he added, he had also seen many poor results. Why aren't they published?

One must be very careful in teaching graduates as well as students, technical methods that are to be applied to these cases and showing them one or two cases of fractured neck of the femur before and after, with a very beautiful result. They go out, and it seems so simple in the hands of expert technicians that it becomes the method of treatment in every hill town in the State. The results of that practice, even now, are not good. There has been a lot of sepsis. There have been a lot of cardiorenal difficulties, a lot of nonunion, protrusion of nails, and all sorts of complications, including deaths under anesthesia, or shortly afterwards. Are these results published? A well known

anesthetist, in Boston, came to Doctor MacAusland and asked: "What am I going to do? Every patient who is nailed takes 2½ hours to operate upon; many die and I cannot stand it. What shall I do?" "Carry on, if I were you," Doctor MacAusland said. "You have a good job."

MacAusland recalled the case of a Hanover Professor who was nailed and was reported as having obtained union, and represented an especially fine result. At the end of three years the man was walking with crutches, having pain, had nonunion, and he could feel the nail rattle around in his hip. There was of course a consultation and Doctor MacAusland said: "It is a nonunion, the nail should be taken out and he should have some type of reconstruction performed." That upset the family and the whole apple cart. There was another consultation. No wonder. Roentgenologic examination was said to have shown trabeculae of bone growing across between the fragments. This was at the end of three years. What nonsense! Operation was imperative, and at operation when Doctor MacAusland made the incision the nail fell out on the floor, and of course there was no sign of union. Why such a report?

Doctor MacAusland said that, although heartily in sympathy with the method, he was not yet convinced that it was the only method, or that the best technic had been developed. He expected to use it more than he has been doing, but was going about it very cautiously. He expressed himself as very definitely against the wholesale teaching of this method, however, as a "cure" for fractured hips because it isn't so, and the statistics that have been reported thus far are not any better, and some not as good, as those obtained by conservative treatment, and many are not true reports!

Some few years ago Doctor MacAusland happened to go on a cruise in which there was a symposium on the treatment of tuberculous joints. The speakers were very carefully selected. They all had the same mind as to what should be done. The only question was, who could get them first and fuse them. He listened to this symposium for one and one-half or two hours, and said that he was very glad to have heard the papers, but felt perfectly sure that Doctor Bradford, his old Professor at Harvard, and an advocate of conservative treatment, must be turning over, face downward, in his grave if he could hear. It didn't seem possible that such treatment could be advocated in such a wholesale manner in every case, and Doctor MacAusland feared that he must say the same thing about fractured neck of the femur. He was definitely opposed to any open operation on patients 80, 85, 90 or 95 years old, that takes so much time and has many pitfalls. He still believed the abduction methods have a very definite place. Certain forms of new traction methods have promise; while he would not care to report his results with this new method as yet, it is certainly as satisfactory as any other procedure and it is distinctly more simple.

Dr. Preston A. Wade (New York, closing) remarked that with one fact, apparently, everyone was in agreement; namely, that subcapital fractures, impacted in the valgus position, unite regardless of what form of treatment is employed. Answering Doctor Gordon's question as to what method he would advocate, since he felt the Whitman method was unsatisfactory, Doctor Wade said he was not advocating any operative method at the present time, because he had not had a large enough series of cases to prove that operative methods were more satisfactory than the older procedures. At the present time many surgeons are trying out various operative methods, as indicated in recent literature—reconstruction operations, pins, Smith-Petersen nails, *etc.* Even Leadbetter states that he is now performing internal fixation in 75 per cent of his cases.

In reporting this series of very poor results following employment of the

Whitman method, Doctor Wade said he meant no disrespect to Doctor Whitman. These cases were treated by 31 different surgeons, and it is helpful to report their results. Doctor Sauer's report of results of treatment in these cases at City Hospital included the cases which received primary treatment from many of the other hospitals in the City. Many of these cases were transferred to the City Hospital during the period of immobilization. They remained at City Hospital for long periods of time, so that end-results were seen.

DR. HERBERT M. BERGAMINI (concluding) quoted the following poem:

THE SMITH-PETERSEN PIN*

A great aunt of mine,
Of age sixty-nine,
Admittedly rather a dreamer,
One day in her flat,
Fell over the cat,
And fractured the neck of her femur.

Doctor Whitman was called,
Who at once was appalled
And made ready to put on a cast.
Although a relation,
A new consultation
I requested without being asked.

It was held the next day,
And on the x-ray
The bone was in fair apposition.
So they seized the old dame,
Strapped her on a hard frame,
As they did in the French inquisition.

Soon that great aunt of mine,
Like an old porcupine,
All tied up with bandage and rope,
Lay bristling with pins
While I for my sins
Supplied the appropriate dope.

In short she got well,
And I shudder to tell
Any facts that are not strictly true;
She's amazingly supple,
And one of a couple
Of tap dancers in a revue.

If you're one of the number
Who, dancing the rhumba
Or after a surfeit of gin,
Fall like my relation,
Without hesitation
Demand a Smith-Petersen pin.

* Author unknown. Original source from a patient who found it somewhere.

# BRIEF COMMUNICATIONS AND CASE REPORTS

## PARATHYROID TUMOR

### REPORT OF TWO CASES

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**Case 1.**—M. C., female, age 46, married, born in Poland, was admitted to Bellevue Hospital October 28, 1932, and was discharged November 10, 1932, with a diagnosis of nodular goiter (adenoma of the thyroid).

*Chief Complaint.*—Swelling on the outer aspect of neck. *Family History.*—Negative as far as goiter was concerned. *Present History.*—This dated back 12 years, when patient noted difficulty in swallowing following childbirth. Five years later she noticed a swelling in the right side of her neck which gradually increased in size. It regularly became enlarged during menstrual periods. There was no tachycardia, no difficulty in swallowing, no nervousness or tremor. Patient had taken iodine off and on for the past year. B. M. R. was within normal limits. Menstrual history was negative. Patient had nine children, seven of whom are living.

*Physical Examination.*—Well nourished woman of 46; good color; weight, 140 pounds; pulse averaged 80; blood pressure, 140/80; lungs, negative; heart, normal in rate and rhythm corresponding with pulse, sounds of good quality, soft systolic murmur at the base. In the region of the right lobe of the thyroid there was a swelling about the size of an orange, somewhat nodular, which moved with swallowing. There was a high pitched bruit heard on the upper surface of the neck. There were no tremors. Urine negative. Wassermann negative.

*Operation.*—November 1, 1932: Under gas-oxygen-ether anesthesia, what was thought to be the right lobe of the thyroid was found involved by a degenerated nodular mass, which was covered by many large veins. It was, however, removed without difficulty. The left lobe of the thyroid was found to be normal. The postoperative diagnosis was nodular goiter. The wound healed by primary union and the patient was discharged from the hospital on the ninth day postoperative.

*Pathologic Examination.*—Doctors Slaughter, Rottino and Symmers: The gross specimen consisted of a mass of tissue 9x6 cm. (Fig. 1). There was a thin transparent capsule, the underlying tissue having a pink color. There were many prominent veins. On section the surface was smooth and homogeneous. Microscopically, the mass was found to be composed almost entirely of oxyphil cells (Fig. 2). *Pathologic Diagnosis.*—Parathyroid adenoma.

The patient was later readmitted to the hospital where roentgenograms of the bones of the skeleton were found to be normal. Blood calcium determination of 10.2 and blood phosphorus determination of 4.2 were within normal limits. Phosphatase was not determined.

Since operation, this patient has remained in excellent health. There has been no recurrence of the tumor. A recent examination showed normal values for calcium, phosphorus and phosphatase.

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The majority of parathyroid tumors reported in the literature have been associated with bone and blood changes. Also, most of the tumors reported are of the clear cell type rather than the oxyphil type as shown in this case.

The case is presented to illustrate a parathyroid adenoma made up almost wholly of oxyphil cells, in whom both the bones and blood chemistry were normal.

**Case 2.**—A. S. C., female, age 53, single, was first examined in July, 1935, at which time a swelling was noted involving the right thyroid region. This had first been noted by the patient two years previously and had been increasing slowly. There was a history of nervousness and fatigue over a period of several years. There had been no loss in weight or any complaint of cardiac disturbance.

FIG. 1. FIG. 2.

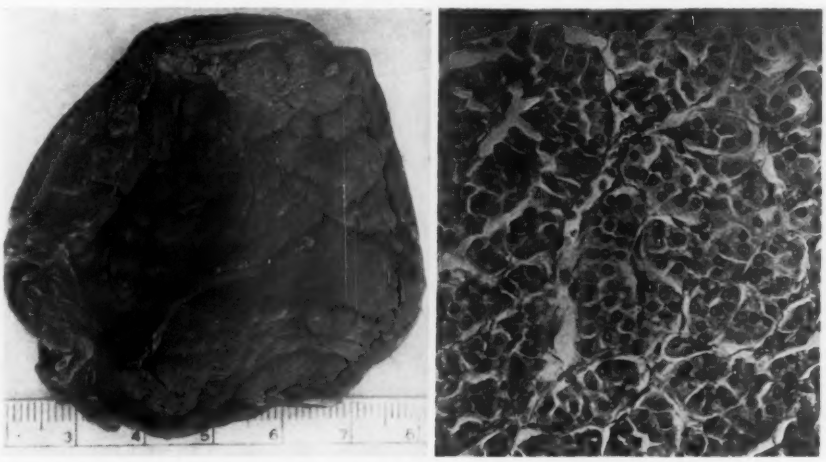

FIG. 1.—Case 1: Photograph of the gross specimen. Note the capsule in left marginal area.
FIG. 2.—Case 1: Photomicrograph of a section of the parathyroid tumor showing the cells well outlined, and the cytoplasm granular and oxyphilic. Note the absence of clear or chief cells.

*Physical Examination* showed a two-inch nodule with a small area of what was thought to be calcification along the inner border, involving what appeared to be the right lobe of the thyroid gland. The weight at this time was 109 pounds (no loss); pulse, 80, and regular; blood pressure, 110/80; B. M. R., —14, which might be considered a low normal. *Diagnosis.*—Nodular thyroid (or adenoma).

A diagnosis of nodular thyroid (or adenoma) was made, and operation for excision of this tumor was undertaken August 24, 1935.

*Operation.*—August 24, 1935: An encapsulated tumor the size of a golf ball, and firmly adherent to the trachea, was found, which apparently involved the right lobe of the thyroid gland. In order to facilitate the removal of the tumor, it was necessary to excise the entire thyroid lobe. On section, the well developed capsule was strongly adherent to the tumor tissue, which was of the consistency of liver and had a yellowish-gray appearance. It was quite evident that the tumor was not a simple thyroid nodule (or adenoma).

*Pathologic Examination.*—Dr. H. R. Muller. *Gross.*—"A spherical mass, about the size of a golf ball. It is covered with a thin fibrous capsule. The consistency is rubbery, but not very hard. The cut surfaces are homogenously pinkish-gray. In one portion is some calcified material. *Microscopic.*—The tumor is composed chiefly of very

FIG. 3A.

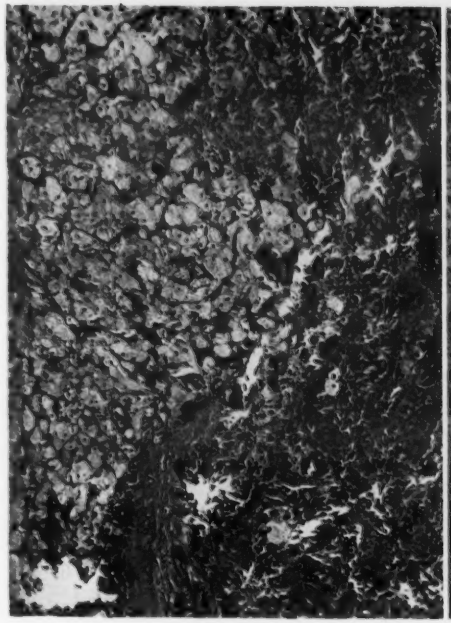

FIG. 3B.

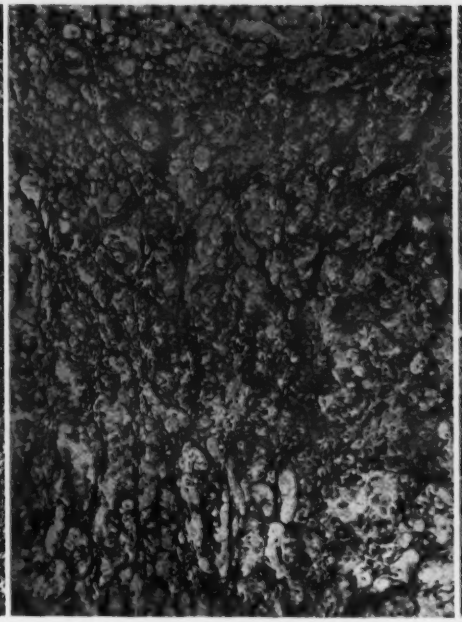

FIG. 4.

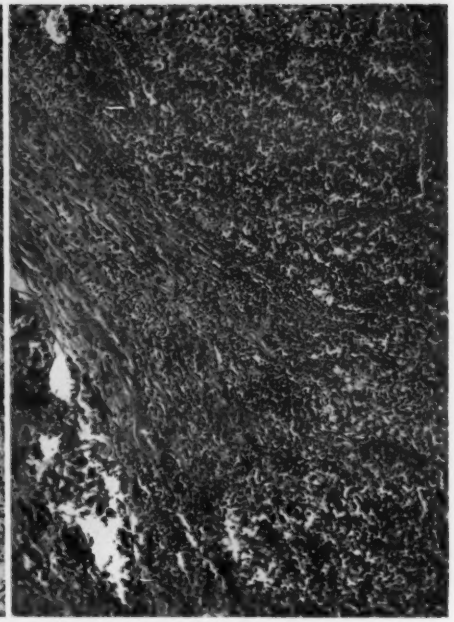

FIG. 3.—Case 2: (A) Showing the lawless arrangement of cells. Note the predominance of clear cells with small nuclei. The darker stained cells are oxyphilic. (B) Note the predominance of clear cells.

FIG. 4.—Case 2: Section of the metastatic nodule attached to the external jugular vein. Note the presence of both clear and oxyphilic cells; also invasion of the capsule.

large cells, with clear, slightly foamy cytoplasm and with a single small round compact nucleus. The outlines of the cells are distinct. Many cells show an area of pinkish material occupying a portion of the cell. A delicate connective tissue reticulum, bearing capillaries, runs around small groups of these cells or acts as a supporting structure. In some portions the cells are small and very acidophilic, but with the same histologic structure. There are also areas of necrosis. The capsule around the tumor varies in thickness, being very thin in places. Outside of the capsule, in one section, is some thyroid tissue consisting of moderately dilated acini filled with colloid and having a low cuboidal epithelium. The structure of this tumor is unusual and suggests an adenoma of the parathyroid. (Fig. 3A and B.) *Pathologic Diagnosis.*—Adenoma of parathyroid."

Calcium and phosphorus determinations, as well as skeletal roentgenograms, were then made and found to be normal.

*Subsequent Course.*—The patient made a good recovery; gained weight; made no complaints, except for occasional painful joints, until August 27, 1937, two years following operation, at which time there appeared a slight fulness in the right side of the neck, and the presence of a small, hard nodule, apparently involving the skin and subcutaneous tissue adjacent to the right external jugular vein on a level with the thyroid cartilage. Physical findings were otherwise negative. There was no loss of weight.

On September 10, 1937, this small nodule was excised under novocain. It was found attached to the external jugular vein. The tissue was friable and gray in appearance.

*Pathologic Examination.*—Dr. E. S. L'Esperance. *Gross.*—"Specimen is an irregular, ragged mass of yellow and grey soft tissue, two centimeters in greatest diameter. *Microscopically,* the sections show a partially encapsulated tumor composed of cells resembling those of the adrenal cortex. The cells, for the most part, are large with round or oval vesicular nuclei. A few have opaque eosinophilic cytoplasm. There is a tendency to glandular arrangement (Fig. 4). *Pathologic Diagnosis.*—This is probably an adenoma of the large cells of the parathyroid. It does not appear to be highly malignant."

*Subsequent Course.*—On December 11, 1937, after roentgenotherapy (four treatments) there was not only a recurrence of the nodule at the site of excision, but there was found another small nodule on a slightly higher level and nearer the median line of the neck, apparently lying superficially in skin and subcutaneous tissue.

On December 31, 1937, roentgenologic examination of the complete skeleton was found to be normal, as was a recent determination of calcium (10.7), phosphorus (3.3), and phosphatase (2.7).

This case is presented as an instance of a malignant tumor of the parathyroid gland; the malignancy based on metastasis rather than on individual cell characteristics. There were no abnormal bone or blood chemistry findings in association with this tumor, which is contrary to some of the reported cases.

DISCUSSION.—DR. EMIL GOETSCH (Brooklyn) felt that Doctor McQuillan's cases represented another confusing syndrome. Here parathyroid tumors were found but there were no associated changes in the bones such as one might be led to expect. The great size of the parathyroid tumors was certainly unusual, the first being about the size of an orange, the second of a golf ball. Doctor Goetsch cited a personal experience he had many years ago with a parathyroid tumor, at least it was so considered after careful studies, as large as a grapefruit, which caused paroxysms of coughing due to tracheal compression. Calcium studies were not made at that time.

In Doctor McQuillan's first case, the histologic section resembled very much the appearance of a thyroid adenoma. In the second case, the tissue

was clearly that of a parathyroid gland. The failure to find incidental bone changes is usually important, and may point to a differentiation in the function of the two main types of cells occurring in the parathyroid gland.

DR. HENRY L. JAFFE (New York) said that a number of tumors, similar to those found in Doctor McQuillan's first case, had been described during the last few years: namely, enormous oxyphil cell adenomata of the parathyroid. The fact that the cytoplasm of the adenoma cells is oxyphilic is offered as the explanation of why these tumors do not demineralize the skeleton; that is, they apparently do not secrete parathormone. It would be of great importance to assay such tumors for the presence of parathormone. One really does not know whether these tumors are parathyroid tumors or not.

DR. ARTHUR S. MCQUILLAN (New York) closing said, in connection with his first case, which Doctor Jaffe discussed, that Keynra, in the British Journal of Surgery, October, 1936, described a case essentially similar, which was made up almost entirely of oxyphil cells, and which evidenced no signs or symptoms of hyperparathyroidism. Keynra cited four other somewhat similar cases and suggested, as did Doctor Jaffe, that possibly these cells are inactive, with low function of secretion and, therefore, one should not expect bone changes.

With regard to malignant tumors of the parathyroid gland, Doctor McQuillan said he had searched the literature and could find only 19 cases of proven malignancy of the parathyroid gland and not all of these were proved by metastases. Diagnosis in some was made on the cell morphology, which is considered difficult, as is the tissue diagnosis of carcinoma of the thyroid gland. All of the 19 cases, except two, did not show any changes in the bones or blood chemistry. The Mayos reported a case, in 1929, of malignancy of the parathyroid gland associated with bone and blood changes. They later reported a case, in 1934, showing the same changes, and in both cases the diagnosis of malignancy was made on cell morphology and not on the presence of metastasis.

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## THE TREATMENT OF ACTINOMYCOSIS WITH THYMOL *

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IN REVIEWING the literature relative to the incidence and methods of treatment of actinomycosis, the following was found: An analysis of 500 cases showed the distribution of the lesions to be: head and neck, 55 per cent; thorax, 20 per cent; abdomen, 20 per cent; and it had occurred in the lower jaw incident to caries of the teeth. Actinomycosis produced the following types of lesions in the region of the appendix: (1) Painful tumor; (2) gangrenous appendix; (3) perforation of the cecum.

Actinomycosis is thought to be contracted from chewing straw, wheat or rye. Various treatments have been tried: Potassium iodide in doses up to 120 gr. or more; wet dressings of copper sulphate ½ to 2 per cent solution; deep

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roentgenotherapy has been of advantage in some instances. Gautier, in France, has developed an electrotechnical method of therapy. He inserts two platinum needles into the infected areas which act as poles between a constant current of 50 milliamperes, and at the same time 10 per cent potassium iodide is injected continuously by the drop method into the mass. The current decomposes this solution into nascent iodine and potassium.

In the present instance, roentgenotherapy was finally decided upon. This had not been considered before because of the possibility of adding sterilization to the other troubles of the patient. Just before the treatment was to be instituted, Dr. H. B. Myers,[1] of Portland, Oregon, published an article describing the use of thymol as a specific for actinomycosis in which he advised adequate surgical drainage of the local lesions and a packing of 10 to 25 per cent thymol in olive oil and tolerance doses of the crystals by mouth to prevent the spread of the disease to other parts of the body.

**Case Report.**—Hospital Chart No. 61057: A. F., white, female, age 20, was admitted to the City Hospital, New York, complaining of discomfort in the right lower abdominal quadrant and the presence of a persistent sinus which opened into the scar of the previous appendicectomy wound.

*Previous History.*—In 1933, the patient, age 15, had been operated upon at the Wyckoff Heights Hospital, Brooklyn, N. Y., for acute appendicitis, but the wound never healed.

*Postoperative Diagnosis.*—Gangrenous appendicitis with local peritonitis. It has not been possible to obtain a pathologic report of the appendix.

*Subsequent Course.*—In December 28, 1936, she was again operated upon at the City Hospital, N. Y., by Dr. Ward Renfrew, and the sinus tract dissected out and an ovarian cyst removed. The wound healed, but one year later, December 28, 1937, she was readmitted with a preoperative diagnosis of acute osteomyelitis of the crest of the ilium. This area was incised and drained. Pathologic examination of tissue removed showed actinomycosis. The wound continued to drain and the patient was given large doses of potassium iodide—up to 120 gr. a day. On March 30, 1937, an abscess in the abdominal wall, in the region of the McBurney incision, was opened, but never healed.

She was first seen by our service on April 1, 1937.

*Physical Examination.*—The patient was thin and emaciated, lying in bed with her right knee drawn up and the right thigh flexed on the abdomen. Weight 70 pounds. She was very apprehensive and frightened. A draining sinus was present at the lower end of her appendix scar, with marked tenderness and induration around it. Over the crest of the ilium there was a broad wound covered with sluggish granulations.

*Operation.*—April 16, 1937: Following a transfusion, the sinus tract leading downward from the lower end of the scar of the McBurney incision was explored. Boggy, granulation tissue was found throughout and the femoral vessels were embedded in it. The tissues bled very freely and the hemorrhage was hard to control. She was subsequently transfused twice because of a severe hemorrhage which occurred at the time of the primary dressing. No improvement followed.

*Second Operation.*—May 13, 1937: This same area was dissected out, only more extensively. It was found necessary to ligate and sever the femoral artery and vein above Poupart's ligament in order to eradicate the involved tissue. The wound extended down to the space of Retzius. An area in the thigh, below Poupart's ligament, was also opened and drained. Notwithstanding the ligation of the femoral vessels, there was never any sign of disturbance in the circulation of the leg. She nearly died during the above operative procedure and had to be transfused immediately postoperatively.

*Postoperative Course.*—The patient became progressively worse. She could not

eat; cried almost constantly from pain; and showed a complete loss of morale. During the last week in May, 1937, her condition had become so serious that roentgenotherapy was considered. However, just at this time the article by Myers[1] appeared, suggesting the employment of thymol. This treatment was immediately instituted, June 2, 1937, and the sinuses were packed with 20 per cent thymol in olive oil. She received 10 gr. of the crystals every other day. Larger or more frequent doses produced a gastro-enteritis. On June 20, 1937, and again on September 8, 1937, areas of infection in the outer side of the thigh were opened and packed. At the first operation it was found that the sinus tract at the lower end of the McBurney scar also opened into the posterior vaginal fornix.

*Results of Thymol Therapy.*—The change in the appearance of the sinus tracts, after packing with thymol, was remarkable. In a very short time they had lost their "drippy," dirty appearance and the granulations became pink, dry and clean. From week to week healing could be seen, especially in the sinus tracts treated with thymol after they had been freshly opened and drained. The temperature dropped immediately. After the operation on June 20, 1937, her progress was very steady. She got up in a wheel chair and then walked with crutches. As soon as she was able, she was sent daily to Occupational-Therapy and allowed out all day in the sun. By September, 1937, the contraction in her knee had disappeared and she walked normally with full range of motion. She has had Alpine light treatments three times a week and transfusions when necessary to keep her hemoglobin within normal range. At present, her sinus tracts are closed except for three. A small one on the anterior surface of the thigh, one on the outer side of the thigh, and the pelvic one which has healed a great deal during the past month. She has a tender, swollen area on the inner side of her thigh which will have to be opened soon, probably an extension of the disease through the obturator foramen along the adductor group of muscles.

The general condition of the patient is good. Her weight is now 107 pounds, a gain of 37 pounds. This new abscess is localized, and certainly the outlook for its eradication by the use of thymol is very much better than it would have been a year ago when her condition was so poor and there seemed to be no prospect of staying the progress of the disease. The red blood count averages about 3,500,000; the hemoglobin between 55 and 75 per cent. The white blood count has averaged about 15,000 with 74 per cent polymorphonuclears. The urine, which during the acute part of her illness showed albumen and casts, has now cleared almost entirely and shows only a faint trace of albumen. This fall, her menstrual periods, which had stopped for seven months, began again and are quite normal.

## SUMMARY

(1) In the case herewith reported, great benefit was derived from the use of thymol by mouth and thymol in oil packed into the wounds after adequate surgical drainage has been established.

(2) No benefit has been derived by the excessive doses of potassium iodide.

(3) The immediate drop in temperature after the administration of thymol, and the later gain in weight, have been evidences of the therapeutic value of thymol.

(4) It is obvious that the patient is not cured, but her condition is certainly vastly better than it was before the administration of this agent.

## REFERENCE

[1] Myers, Harold B.: Thymol Therapy in Actinomycosis. J.A.M.A., **108**, 1875, May 29, 1937.

DISCUSSION.—DR. THOMAS H. RUSSELL (New York) cited the following case: A white, Jewish male, age 28, was operated upon in Florida for a ruptured appendix. He felt well for a few weeks, but then his temperature rose and a diagnosis of subphrenic abscess was made and the abscess drained. He improved, but four months later because of a temperature of 102° F. the subphrenic abscess was reopened and drained. Following this his temperature did not come down to normal, and he was admitted to New York Post-Graduate Hospital for reexploration of the subphrenic abscess. At operation, January 20, 1937, the subphrenic area was explored. No pus was found but a moderate amount of necrotic material was removed from the sinus and subphrenic area. Since then there has been a small amount of discharge from the tract. Scrapings from the sinus tract revealed purulent exudate with associated colonies of actinomycosis. Gastro-intestinal roentgenologic studies, Februrary 10, 1938, revealed a rather narrow, constricted terminal ileum, somewhat segmented, suggesting chronic hypertrophic changes in this segment. Roentgenograms of the lung showed subphrenic pathology, moderate lung retraction on the right side with fluid at the right base, and a partial hydropneumothorax. For the past six days the patient has been treated with thymol, taking 2 Gm. daily by mouth, and the sinus being irrigated with 15 per cent thymol solution in olive oil.

DR. ALLEN O. WHIPPLE (New York) said that he had had one patient who was proving a very interesting problem, who had a very diffuse distribution of actinomycotic lesions. The difficulty was that he had had a number of therapies instituted, as is so often the case in these desperate lesions, and it was very difficult to determine just which therapy had had the most pronounced effect. Thymol regimen would have been pushed much more vigorously had the patient tolerated it better. Tolerance should be determined, as some patients are quite intolerant to the large doses employed. There are several ways of administering thymol. Taking it by mouth upsets some patients so that they cannot take food. The case just referred to was up and about with a new focus, and he thought the excellent result largely due to the indefatigable and constant attention of the house surgeon who had really done a remarkable piece of work in keeping after the patient. Undoubtedly thymol has a very real place in the therapy directed against this condition.

DR. M. STANLEY-BROWN (closing) said that in Dr. H. B. Meyers' description of the use of thymol there was one case included, in the six reported, which did not turn out favorably because the patient could not tolerate the thymol by mouth. The local lesion cleared up but the patient developed actinomycosis of the lung from which he eventually died. Where the thymol is tolerated it seems to work very well. Doctor Meyers stressed the point that the affected areas must be opened, afforded adequate surgical drainage, and packed with thymol in olive oil to give the best results. The patient presented weighed 70 pounds when first seen, and had been in the hospital for several months with no change. After thymol was instituted she gained weight and her temperature dropped; it would seem to be the effect of the thymol therapy that brought about the improvement. She tolerated 10 Gm. every other day, but if this amount was increased some difficulty was experienced.

# REVERSED COLLES' FRACTURE

LEEMAN E. SNODGRASS, M.D.

PHILADELPHIA, PA.

THERE is a discussion in the literature, Webb and Sheinfeld,[1] Raymer,[2] Bettman and Tannenbaum,[3] Wood,[4] Greeley and Hobart,[5] on the question of whether or not reversed Colles' fracture can be reduced and held, without resort to open operation. It has been of interest to study the photographs published by these authors, in conjunction with the five cases, shown in Figures 1, 2, 3, 4 and 5, from the Episcopal Hospital records.

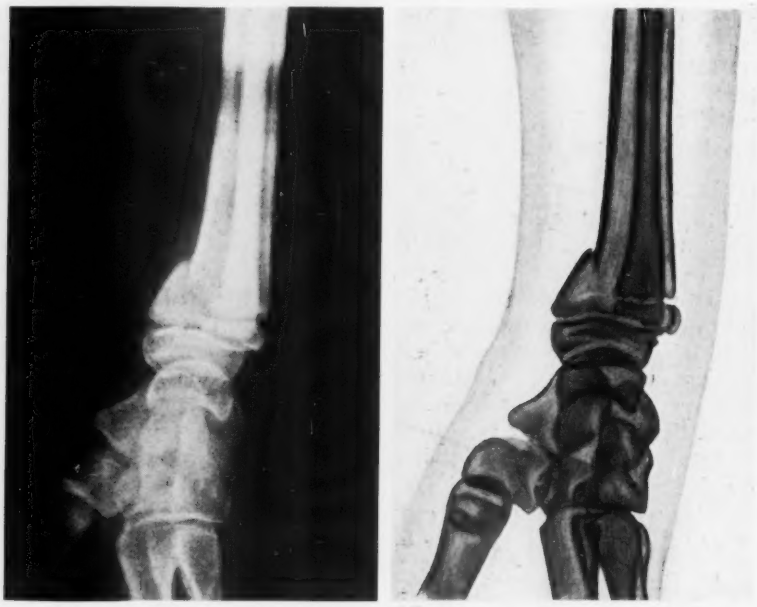

FIG. 1.—B. R., age 11. Greenstick fracture in distal end of diaphysis with slight palmar displacement of fragment. A true reversed type.

It is important for the proper understanding of this lesion to realize that we are dealing with two rather distinct conditions. Some of them are true reversed Colles' fractures; others are anterior fracture-dislocations of the wrist. The true reversed Colles' fracture is very well shown in Greeley's and Hobart's[5] Figure 7B, in which dislocation is absent, and in which the fragment is displaced at an angle to the shaft. The fracture-dislocation type is equally well shown in their Figure 1, in which the wedge-shaped fragment from the lower lip of the articulating surface of the radius is approximately parallel to the anterior surface of the shaft. It has been forced proximally away from the line of the radiocarpal joint. Comminution of the fragment may produce an intermediate type, as shown in Figure 4.

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This fracture-dislocation bears some resemblance to fracture of the dorsal lip of the radius. Both of these lesions represent fractures of overhanging bony lips. The difference in the two is to be found in the fact that the dorsal lip of the radius overhangs the radiocarpal joint, while the anterior lip is approximately flush with the articulating surface, but overhangs the anterior surface of the shaft of the radius. These two lips protrude approximately at right angles to each other.

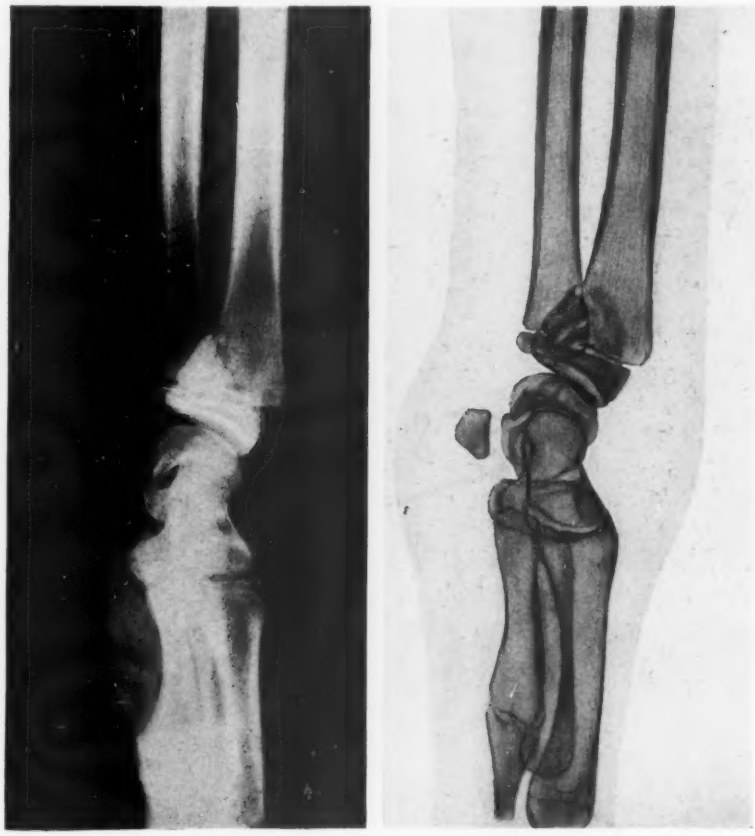

FIG. 2.—D. O., age 13. Epiphyseal separation anteriorly. A true reversed type.

In fracture of the anterior lip, the integrity of the joint is destroyed, and the carpus is easily displaced along the anterior surface of the shaft. In fracture of the dorsal lip, it is rare for dislocation to occur, because of the intimate relationship which the strong flexor tendons bear to the anterior surface of the carpus. These tendons turn sharply about the lower end of the radius in dorsal flexion, and unless the entire lower end of the radius is fractured and displaced, the carpus cannot be forced dorsally while in dorsal flexion. A dislocation of the carpus in dorsal lip (Barton's) fracture would,

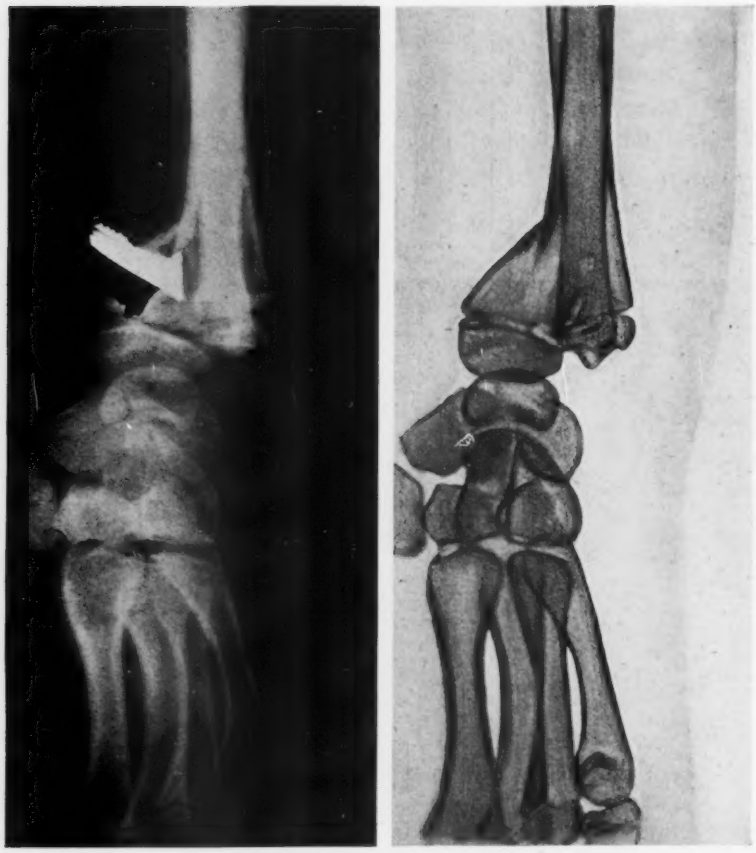

FIG. 3.—E. W., age 13. Epiphyseal separation anteriorly. A true reversed type.

FIG. 4.—I. B., age 67. An intermediate type. The anterior lip of the radius is broken off, but the fracture line also passes completely through to the dorsum. This intermediate type is formed by the comminution present.

of necessity, be at a right angle to the tendons. In the anterior fracture-dislocation, the dislocation is in line with the tendons and the fragment can slide along them.

The closed reduction and fixation of fracture-dislocation is conceivably more difficult than it is in the case of the true reversed Colles' fracture.

FIG. 5.—M. G., age 45. Anterior fracture-dislocation of the wrist. The fragment is parallel to the shaft of the radius. The anterior lip of the radius is fractured and driven proximally.

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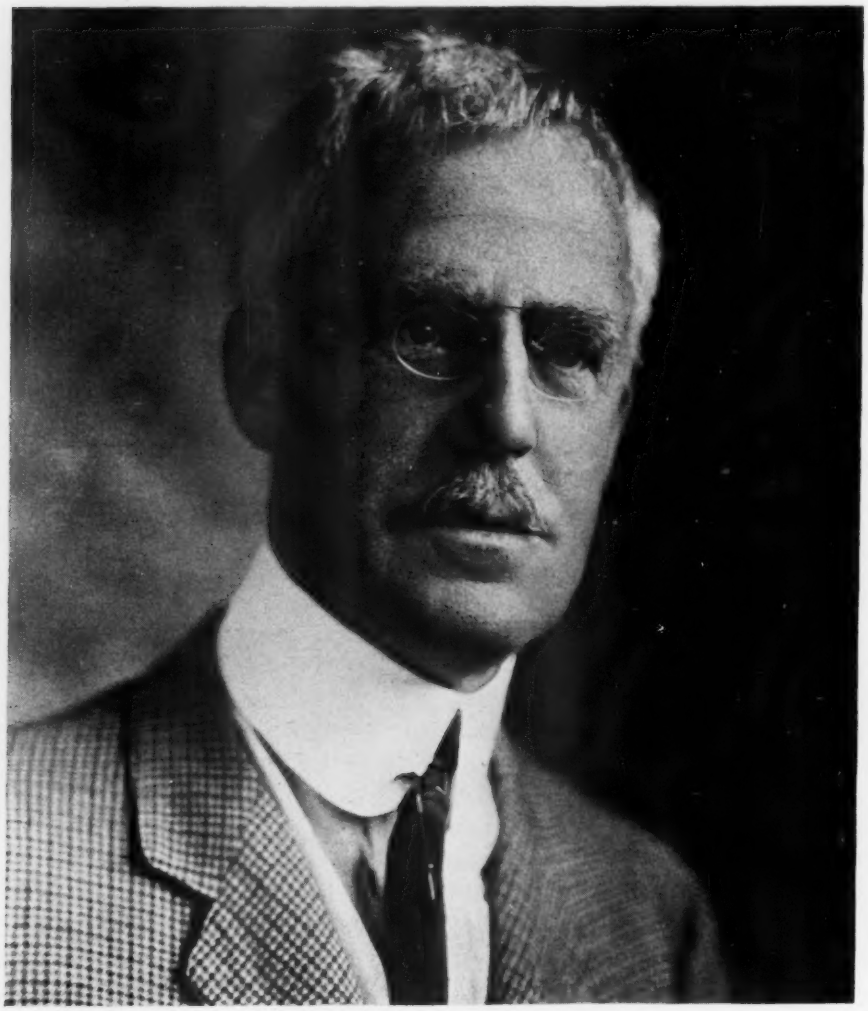
# MEMOIR

## EDWARD MARTIN

### 1859–1938

DOCTOR EDWARD MARTIN, the youngest son of Jonathan Willis and Malvina Regester Martin, was born in Philadelphia August 14, 1859, and died in the same city, March 17, 1938. His preliminary education was ob-



EDWARD MARTIN, M.D.

tained in the public schools of this city and for a short period at Central High School. He attended Swarthmore College four years, ranking first or second in all classes and was elected to Phi Beta Kappa. He was graduated in 1878, Magna cum laude, obtaining the degree of A.B. at that time and later (1882) A.M. and (1920) D.Sc. from this college. He was graduated from the medical school of the University of Pennsylvania in 1883, from which institution he later received the degree of LL.D., and served as assistant to Dr. D. Hayes Agnew and later to Dr. J. William White. For a few years he practiced medicine and then devoted his time to working in the fields in which he ultimately specialized—genito-urinary and general surgery. He was a prolific writer, being the author of several authoritative text-books on surgical subjects, sections for medical systems, editor of the University Medical Magazine and Surgical Editor of The Therapeutic Gazette, as well as the author of innumerable papers read before various medical societies and associations. He was a true teacher, with the ability to so present his thoughts that he indelibly impressed them upon his listeners.

His loyalty and affection to Swarthmore College and the University of Pennsylvania were outstanding—nothing that he could do ever seemed to be enough, in his estimation, to repay these two institutions for what they had done for him. He worked indefatigably for them in every way that he could. His motto was "The Best Is Yet to Come"; and with this in view he worked and planned to the last for work in the Edward Martin Biological Laboratory at Swarthmore, which laboratory was really a tribute to the lasting friendship existing between Mr. Fred M. Kirby and Doctor Martin, and was made possible by Mr. Kirby's generous million dollar gift for this purpose.

Another permanent memorial to this friendship was the Kirby Fund, founded by Mr. Kirby for the benefit of students who needed financial aid to complete their medical course. It was instituted after Mr. Kirby heard Doctor Martin tell of a student who tried to work his way through medical college and died in the effort. Under this Fund students were enabled to borrow such money as they required, paying it back to the Fund as soon as they became able to do so. He helped financially, and through his influence obtained scholarships for more than one young man—and these men have since achieved success in the medical and other professions, thanks to his efforts and encouragements.

He was a pioneer in many lines—prior to the Wright brothers' achievements, he and Dr. Paul Chamberlin conducted a number of experiments to prove air flight possible. He also was interested in the Eastman Kodak Company from a teaching standpoint and made several trips to their plant in Rochester, N. Y., to supervise the taking of educational films—one of which dealt with appendicitis—to be used in the medical schools. He was a pioneer in the use of the lantern slide and later movie films for student teaching.

He was the first man in the University of Pennsylvania Hospital to use

morphia after an abdominal operation; and to Doctor Martin we owe the deep breathing exercises to prevent postoperative pulmonary complications, which practice has withstood the test of time and is today used even more commonly than $CO_2$ inhalations. He early practiced the "no-touch" or instrument technic in surgery, using white cotton gloves as telltales on the offender. Gentleness in the handling of tissue was his continual admonition.

In all things, Doctor Martin believed in preparedness, and this, with his ideas on loyalty, honesty of purpose, kindliness, tactfulness and consideration in the handling of his patients, constituted his creed. He was honest with himself and others in all his dealings, a strict disciplinarian, yet most charitable to the failings of others.

Doctor Martin had many allied activities in public enterprises. It was through his efforts that typhoid fever was stamped out during the Spanish-American War; he was Director of Health and Charities in Philadelphia under Mayor Weaver, from 1903 to 1905; Commissioner of Health in Pennsylvania, from 1918 to 1923, under Governor William Sproul (a classmate of his at Swarthmore); he inaugurated the Traveling Dental and Prenatal Clinics; did special work regarding syphilis and gonorrhea; instituted the use of toxin-antitoxin for preschool age children, and of antitoxin early in diphtheria; was responsible for schools of instruction held one week each summer for State Inspectors. The Philadelphia Health Council and their good work against tuberculosis are the result of his efforts.

Since 1885, he was a member of the Board of Managers of Swarthmore College; a member of the Board of Education, since 1911, and its President at the time of his death. He was elected Clinical Professor of Surgery at the Women's Medical College of Pennsylvania, in 1902, resigning after several years. He was Professor of Clinical Surgery at the University of Pennsylvania, 1903 to 1910, and John Rhea Barton, Professor of Surgery at the same institution, from 1910 to 1918. He also served the University of Pennsylvania as Professor of Surgical Physiology. He was Chief Surgeon to the University of Pennsylvania and the Howard Hospitals, from 1910 to 1918, and consulting surgeon to Bryn Mawr Hospital, Norristown State Hospital for the Insane, State Hospital for the Insane at Wernersville and the Philadelphia General Hospital.

He was a Fellow and Regent of the American College of Surgeons and former president of the Clinical Congress of Surgeons, a member of the American Philosophical Society, American Surgical Association, Clinical Society of Surgeons, American Association of Genito-Urinary Surgeons, and of several foreign surgical societies, as well as the local state and county organizations. Incidentally, he was a member of the Philadelphia County Medical Society for 54 years and received a framed certificate to that effect which he prized very highly. In 1935, Temple University conferred on him the degree of D.Sc.

Doctor Martin's war record covered a period of many years, he having been a Reserve Officer for some years and in the O.R.C. several years be-

fore the World War. His active service in the World War began in Philadelphia January 1, 1918, although he had been a member of the Medical Section, Council of Defense, from the beginning of the war. He was lieutenant, major, lieutenant colonel, colonel, reserve officer of the World War—364th Medical Regiment, O.R.C., U.S.A.; Regimental and Brigade Surgeon, Spanish-American War; Surgeon and Major in Third Regiment National Guard of Pennsylvania for 12 years in the early nineties; Director of School of Surgery, Fort Oglethorpe, Ga.; also Chief of Surgical Section of Hospital. He was the first director of Base Hospital No. 20 of the University of Pennsylvania; however, he was soon called back to Washington, being succeeded by his assistant, Dr. J. B. Carnett, who carried it to its fruition. Doctor Martin was Chairman of States Committee, School of Surgery, University of Pennsylvania, for Medical Officers of the Army; Director of Professional Service and Surgical Chief, Walter Reed Hospital; Chairman of States Committee, Advisory Board Medical Section Council of National Defense. He was honorably discharged from the service January 27, 1919.

Athletics always aroused his greatest enthusiasm. He was on a winning crew of the University of Pennsylvania in the eighties, played tennis, fished, and in later years was an ardent golfer. At the close of the football season he acted as host to the entire football squad of Swarthmore College, giving a traditional dinner at The Lamb's Tavern with the main feature on the menu being a suckling pig with a red apple in its mouth!

For a number of years, beginning in 1903, Doctor and Mrs. Martin entertained children from the city slums at Valley Farm, a short distance from their Orchard Farm. These youngsters were exceptionally well cared for during their stay and were returned to their homes after a vacation of a week or two; Girl Scouts have occupied a part of Doctor Martin's farm for several years, his cousin presiding as hostess. For the past 20 years, Doctor Martin spent his winters in Florida, his summers at Orchard Farm, Media, Pa., and about a month at his home in Graefenburg Springs in the beautiful South Mountain part of the Blue Ridge chain. There were many deer here and he planned to give this estate to the State as a game preserve after the death of Mrs. Martin and himself.

He was a member of the Jury, Rittenhouse, Pine Valley Golf, Rolling Green Golf and Seaview Golf Clubs.

Endowed with a keen, quick sense of humor, Doctor Martin, with his six foot one inch straight soldierly carriage and dark auburn hair, was much in demand among his friends and professional colleagues, excelling alike as speaker, toastmaster or host—he was never at a loss for the appropriate word or expression in any situation. He was extraordinarily clever as a correspondent and always kindly, sympathetic and helpful to those who were fortunate enough to come in contact with him. He was quick to sense any possible talent in those in whom he was interested and would do all in his power to bring such talent to perfection. The death of his wife last fall after many years of companionship was a great loss to him.

And so lived a great man—a true servant of the public—whose virtues often escaped the superficial observer as is true of so many of the world's kindest and most helpful members whose sincerity and naturalness lead them to hide their real selves behind a "smoke screen" to escape embarrassing acclaim. Of no one was this more true than of that Hicksite Quaker—Edward Martin.

ELDRIDGE L. ELIASON.

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